DOCTOR OF CLINICAL PSYCHOLOGY (DCLINPSY)

Doctorate in Clinical Psychology: Main Research Portfolio

1) Critical Review of the Literature: A systematic review of the psychosocial outcomes of sibling bereavement in childhood and early adulthood; 2) Service Improvement Project: Improving the process for referrals for psychological input: how recovery team employees make decisions to refer; 3) Main Research Project: Guilt and shame in perinatal death: a comparison of childless women and women who have children.

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A Systematic Review of the Psychosocial Outcomes of Sibling Bereavement in Childhood and Early Adulthood

Critical Review of the Literature

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Target journal: Journal of Death and Dying (for details see Appendix 1.1)

This journal takes brings insight into terminal illness; the process of dying, bereavement, mourning, funeral customs, suicide and draws significant contributions from the fields of psychology, sociology, medicine, anthropology, law, education, history and literature.

Abstract

This review summarises studies using validated instruments to measure short and long-term psychosocial outcomes among siblings bereaved in childhood and early adulthood. A systematic search was conducted of PubMed, PsychINFO and Scopus databases for articles relating to psychosocial outcomes of sibling bereavement for people aged 0-25 years. Results yielded 1,192 references, generating articles reporting 18 unique studies meeting inclusion criteria. Quality assessment indicated that most studies had high risk of bias.

Consistent with the literature, bereaved siblings had a higher risk of poor psychosocial outcomes in the first two years, but most recovered in the long-term. In the short-term, bereaved siblings experienced similar levels of distress to parentally bereaved children. Factors associated with poor psychosocial outcomes were summarised in the context of an integrative risk factor framework. There was some evidence that younger children are affected more than older children and girls may be more affected in the long-term. Parental distress, lack of communication and lower social support may be associated with poorer outcomes but the high risk of bias in studies means that conclusions are tentative.

Clinicians should be aware that sibling bereavement can affect children's emotions and behaviour and of the importance of communication and of the relationship between parental and children's distress. Future research is needed into the effects of relationship breakdown and different family structures and the impact of social media support groups.

Key words: Childhood, adolescence, bereavement, sibling, death, grief psychosocial outcomes, systematic review

Introduction

Children usually experience despair, grief and sadness as a result of the death of a close family member (Dowdney, 2000). Studies have indicated that in the short and medium-term bereavement in childhood and adolescence is a risk factor for distress and dysfunction, with adverse outcomes including depression, anxiety (Kaplow, Saunders, Angold, & Costello, 2010), sleep disorders, somatic problems (Kalter et al., 2003), behavioural problems and difficulties making friends (Fauth, Thompson, & Penny, 2009; Worden & Silverman, 1996). Within two years, most bereaved children appear to recover to normal levels of functioning, but 15-20% experience longer term distress (Currier, Holland, & Neimeyer, 2007; Dowdney, 2000; Worden & Silverman, 1996).

This pattern, whereby most children experience short-term problems and recover, but a minority experience longer term problems, is consistent with the trajectory of grief in adults (Bonanno, 2004; Bonanno et al., 2002). For adults, Stroebe, Folkman, Hansson, and Schut (2006) have developed an integrative risk factor framework for understanding individual differences in bereavement outcomes based on the dual process model (DPM) of grief which suggests that grieving is a process of oscillation whereby the bereaved moves between confronting their loss (loss-oriented behaviours) and avoiding it to deal with everyday tasks (restoration-oriented behaviours) (Lazarus & Folkman, 1984; Schut & Stroebe, 1999; Stroebe & Schut, 2010).

Stroebe et al. (2006) propose that understanding risk factors requires analysis of three elements: (1) stressors, which could be loss-oriented, (e.g., related to closeness of relationship with the person who has died or to cause of their death) or restoration-oriented (e.g., the death meaning that there is less income within the family); (2) interpersonal and intrapersonal resources e.g. social support and attachment style; and (3) coping and appraisal processes, e.g. cognitive and behavioural mechanisms in appraising the two elements above and their emotional regulation (see Figure 1.1 below). Although this model has not been empirically tested with children, its potential relevance to child bereavement has been recognised (Stokes, Pennington, Monroe, Papadatou, & Relf, 1999).

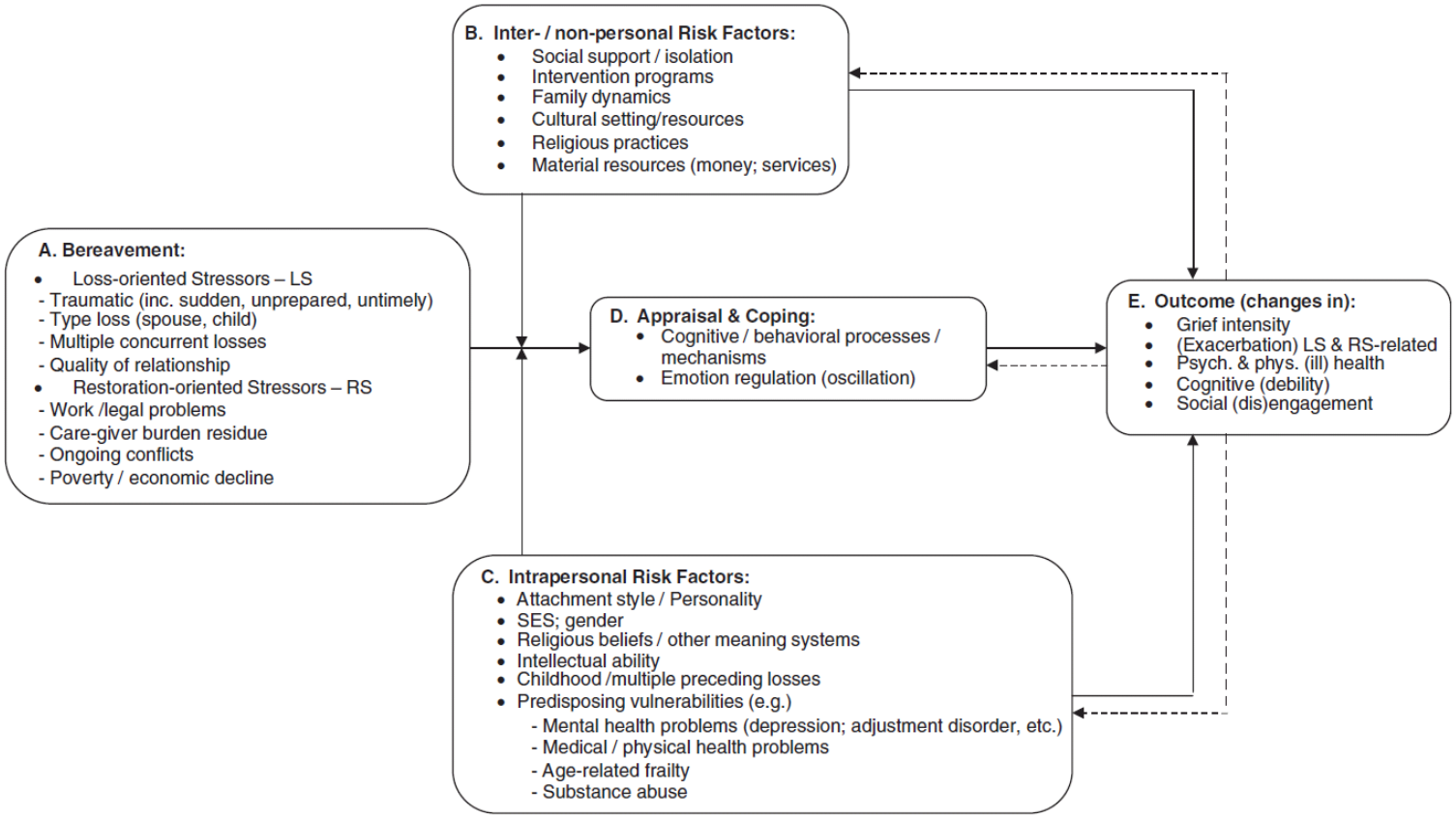

Figure 1.1. Integrative risk factor framework for the prediction of bereavement outcome. Reprinted from "The prediction of bereavement outcome: Development of an integrative risk factor framework" by Stroebe et al. 2006, *Social Science and Medicine*, 63(9), p.2444. Copyright 2006 by Elsevier. Reprinted with permission.

Currier et al. (2007) found in a meta-analysis of grief interventions for children that treated children did no better than bereaved children who did not participate in grief therapy. However, they also found that interventions were valuable when children are already exhibiting difficulties and have a genuine need for treatment. Since this review was published, a randomised control study found that a family intervention with parentally bereaved children was effective for those with problematic grief (Sandler et al., 2010). This means that naming the psychosocial outcomes for children who experience specific types of bereavement and investigating the factors associated with more prolonged or severe grieving processes may enable more targeted interventions, benefiting those likely to have longer-term distress.

Sibling Bereavement

The impact of the death of a sibling in childhood and early adulthood (when the birth family is usually the primary family unit) is an area of increasing interest in bereavement research. Siblings contribute to cognitive and emotional development and adjustment, both directly through their encounters with each other and indirectly as their own relationships with their parents are influenced by their sibling's impact on parents (Brody, 2004; Dai & Heckman, 2013; Gass, Jenkins, & Dunn, 2007; Ruffman, Perner, Naito, Parkin, & Clements, 1998). The consequences of sibling bereavement in childhood were studied by Cain, Fast and Erikson as early as 1964 and there are several qualitative studies which explore children's experiences of sibling bereavement in the context of continuing sibling bonds and changes in parental relationships (e.g. Foster et al., 2009; Hogan & DeSantis, 1992; Packman, Horsley, Davies, & Kramer, 2006). However, until relatively recently, quantitative research into bereavement in childhood and adolescence had predominantly focused on the impact of parental death, with few studies examining the impact of sibling death (Dowdney, 2000). In the last decade, several studies have explored the short and long-term effects of sibling bereavement and factors associated with such outcomes (e.g. Bolton et al., 2016; Eilegard, Steineck, Nyberg, & Kreicbergs, 2013b; Gerhardt et al., 2012; Morris, Gabert-Quillen, Friebert, Carst, & Delahanty, 2016). Therefore, a review of the current research is timely.

Aim of Review

The aim of the review was to offer a systematic summary of studies investigating psychosocial outcomes (e.g. mental health outcomes, education and employment outcomes) associated with sibling bereavement in childhood and the related factors. The evidence in relation to factors affecting psychosocial outcomes was summarised within Stroebe et al.'s (2006) framework. The focus of the research was bereavement in the context of sibling relationships and so studies whose primary focus was the investigation of psychosocial outcomes resulting from deaths occurring before the birth of sibling, perinatal deaths and sudden infant death syndrome deaths were excluded. In addition, studies which primarily focused on the psychosocial outcomes of deaths from homicide and suicide were also excluded because of their focus on the complications arising as a result of the type of death rather than the sibling relationship (e.g. Dyregrov & Dyregrov, 2005; Pfeffer et al., 1997; Pitman, Osborn, King, & Erlangsen, 2014). However, studies were included where they reported on the psychosocial outcomes of sibling bereavement of an Index Child which included these causes of death (i.e. occurring prior to the birth of a sibling, perinatal deaths, sudden infant death syndrome, suicide and homicide) provided the majority of deaths did not fall into these categories. This was because the focus was not specifically on the consequences from these types of death, but on sibling bereavement generally. The following questions were addressed:

1. What are the short-term and long-term psychosocial outcomes of the death of a sibling for children and young adults aged between 0-25 years?

2. What factors are associated with individual differences in psychosocial outcomes?

Method

Search Strategy

Electronic databases (PubMed, PsychINFO and Scopus) were searched with no restrictions of publication year. The following combinations of keywords were used to identify eligible publications: Grief (Mesh term)/grief/Bereavement (Mesh term)/bereavement AND Siblings (Mesh term)/brother/brothers/sister/sisters. Reference sections of included studies and narrative reviews were screened to identify additional

studies and a cited reference search was conducted for each included study to capture other relevant studies. The last search was performed on 18 August 2016 (Appendix 1.2).

Selection of Literature

The PECO framework used in this review defining the (P)opulation, (E)xposure (C)omparison and (O)utcome of interest was as follows:

- *P* Individuals bereaved before 25 years of age.
- *E* Death of a sibling.
- *C* Statistical examination of the relationship.
- *O* Psychosocial outcome measured by validated instrument or clinical records.

Inclusion criteria. Studies were included if they (a) included a population of children and young adults bereaved by the death of a sibling (the "Index Child") between the ages of 0-25 years; (b) examined psychosocial outcomes through comparison with normative data and/or a control group and/or analyses of predictive variables for psychosocial outcomes for the bereaved group; (c) included quantitative data and used a validated outcome measure or third-party source (e.g. hospital records); (d) were published in English; and (e) were primary research published in a peer-reviewed journal.

Exclusion criteria. Studies were excluded if: (a) the majority (i.e., over 50%) of the deaths of the Index children were reported as having occurred in the following circumstances: (i) siblings had not been born when the Index Child died; or (ii) the Index Child died within 28 days of birth, or died of sudden infant death syndrome; or (iii) the Index Child was murdered or had committed suicide; (b) the study used qualitative methods; (c) the study was a single case study or review.

Process of Literature Search

References were imported into Endnote and duplications were removed. Titles and abstracts were studied to determine selection for full-text reading and resulting full texts were studied to determine inclusion.

Inter-rater reliability. Twenty percent of titles and abstracts (*N*=139) were randomly selected and assessed by a second researcher to pilot the exclusion criteria. Inter-rater agreement was excellent with a Cohen's kappa of 93%. Although there was some disagreement, the second researcher had not excluded any abstracts initially included by the first researcher and so the full-texts of all abstracts and titles selected by the first researcher were reviewed. The second researcher also assessed 20% of the full-text articles reviewed (*N*=12). Inter-rater agreement was perfect with a Cohen's kappa of 100% of the full-text articles and therefore no disagreements needed to be resolved.

Data collection and quality assessment. Information was extracted on population, design, comparison groups, follow-up and results. Multiple papers from the same study were linked.

Quality of studies. There was variation in methodology of the studies reviewed and so the Newcastle-Ottawa Scale (NOS) (Wells et al., 2007) for systematic reviews of observational studies was adapted with reference to guidelines of prognostic studies (Hayden, Côté, & Bombardier, 2006). Quality was assessed according to (1) representativeness of cases; (2) control group and selection; (3) assessment of associated factors, e.g. whether by retrospective report; (4) important confounders accounted for in the study design, e.g. age of participant at time of death, time since death, whether sibling in same family is included in the analysis; (5) outcome assessed by validated measure(s); (6) clarity of hypotheses and associated analyses. All criteria were measured on a 0-2 scale (maximum score 12).

Results

Literature Search

Figure 1 illustrates the flow of studies through the review. The search identified 693 references after removal of duplicates yielding 24 papers reporting 18 unique studies for the narrative synthesis, including one study identified through review of reference lists.

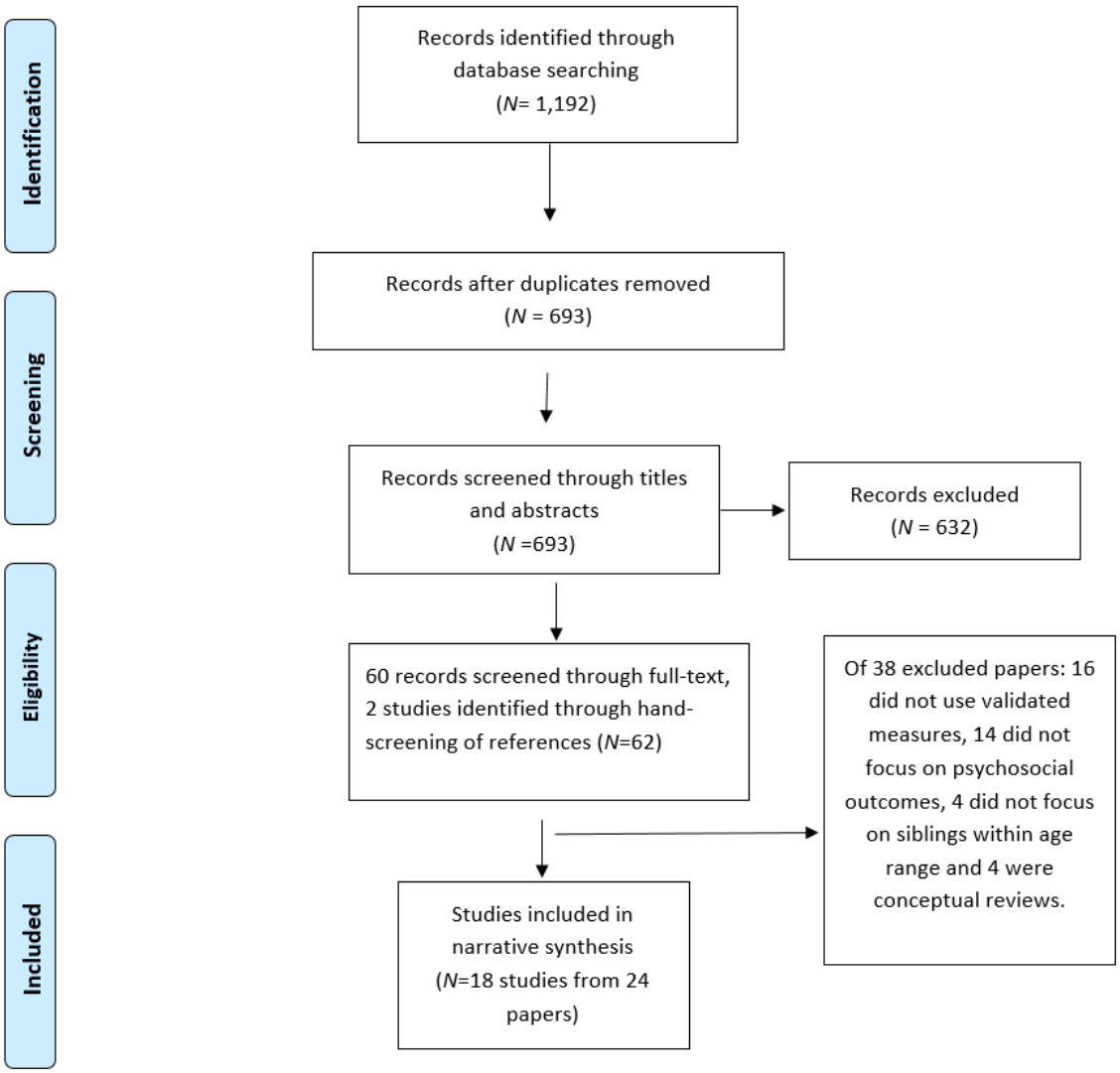

Figure 1.2. Flow diagram of study selection and review

Description of Studies

Fourteen studies were conducted in the United States. The remaining studies were performed in Canada (1), Sweden (1), Holland (1) and the United Kingdom (1). There were three cohort studies, eight studies recruited from hospital records and seven studies recruited from self-help groups or researcher's professional contacts. Studies investigated the effects of sibling bereavement due to cancer (k = 7), accidental death (k = 1) and varied

or unreported causes of death (k = 10). The studies reported on 9701 bereaved siblings, 8864 from large cohort studies. Psychosocial outcomes investigated included mental health outcomes, emotional and behavioural difficulties including grief, social relationship measures and life style measures (e.g. marital status and employment measures). Nineteen validated measures were utilised to measure psychosocial outcomes (Appendix 1.3). Six validated instruments were utilised to measure factors associated with psychosocial outcome (Appendix 1.4). Methodological quality ratings for all studies are outlined in Table 1.1 (maximum score 12). All studies are summarised in Table 1.2.

Table 1.1.

Quality assessment of studies included in review (see Appendix 1.5 for details of reasoning)

Study and number of participants	***Selection bias***	***Control group***	***Associative factors (bias where retrospective reporting)***	***Confounds (e.g. from same family, no pre-testing)***	***Outcomes***	***Statistics***	***Quality Total (max 12)***
1. Applebaum and Burns (1991) (N=20)	0	0	1	1	2	2	6 (50%)
2. Balk (1983a); Balk (1983b) (N=33)	0	0	1	1	1	1	4 (33%)
3. Birenbaum et al. (1989) (N=61)	1	0	1	1	2	1	6 (50%)
4. Bolton et al. (2016) (N=7243)	2	2	2	2	2	2	11 (92%)
5a. Davies (1988) (N=55)	1	0	0	1	1	1	4 (33%)
5b. Worden et al. (1999) (N=75)[a]	1	1	1	1	1	1	6 (50%)
6.Demi and Gilbert (1987) (N=18)	0	0	1	0	0	2	3 (25%)
7. The Swedish Study 7a. Eilegard et al. (2013b) (N=174)[a]	2	2	2	1	1	2	10 (83%)
7b. Wallin, Steineck, Nyberg, and Kreicbergs (2015)[a]	2	0	0	0	1	1	4 (33%)
7c. Eilertsen, Eilegard, Steineck, Nyberg, and Kreicbergs (2013)[a]	2	0	0	0	1	1	4 (33%)
7d. Sveen et al. (2014)[a]	2	0	2	1	1	1	7 (58%)
7e. Lövgren, Jalmsell, Eilegård Wallin, Steineck, and Kreicbergs (2016) [a]	2	0	0	0	1	1	4 (33%)
8. Fletcher, Mailick, Song, and Wolfe (2013) (N=1621)	2	2	2	1	1	2	10 (83%)
9 Gerhardt et al. (2012) (N=105)	1	2	2	1	2	2	10 (83%)
10. Hogan (1988) (N=40)	0	0	2	1	1	0	4 (33%)
11. Hogan and Greenfield (1991) (N=127)	1	0	1	0	1	1	4 (33%)

12. Martinson et al. (1987) (N=29)	0	0	1	1	1	1	4 (33%)
13. McCown and Pratt (1985) (N=66)	1	0	1	1	1	1	5 (42%)
14. Morris et al. (2016) (N=62)	1.	0	1	2	1	2	7 (58%)
15. Mulhern et al. (1983) (N=46)	1	0	1	0	1	2	5 (42%)
16. Pettle Michael and Lansdown (1986) (N=28)	1	0	1	0	1	0	3 (25%)
17. Rosenberg et al. (2015) (N=56)	1	0	1	1	1	1	5 (42%)
18. Stikkelbroek et al. (2016) *(*N=15)	1	2	1	1	1	2	8 (67%)

[a] Quality assessments were conducted where papers reported analyses of a study since quality varied according to the factors and outcomes reported.

Table 1.2.

Summary of included studies in alphabetical order.

Author	*Population (N)* *Sibling age* *Cause of death of Index Child* *Time since death*	*Study design and comparison group*	*Associative Factors*	*Measure of sibling's adjustment (validated instrument)*	*Findings*	*Quality rating*
1 Apple-baum and Burns (1991)[a]	N=20, 9f (45%), 11m (55%). Recruited from self-help groups. Age at time of study: 3-23 yrs (*M* = 15.1yrs, *SD* = 5.8 yrs). Cause of death: accident = 10 (50%) homicide = 10 (50%).[a] Time since death: 4-77 months (*M* = 35 months *SD*=24).	Cross-sectional Comparison of siblings according to cause of death	Parental PTSD Sibling's age Time since death Closeness in age to sibling	Trauma **CRI** Childhood post-traumatic stress disorder (PTSD) (parent report) Child self-report of PTSD	No effect of cause of death on PTSD symptoms.[a] 45% of siblings had PTSD. Parental PTSD positively correlated with siblings' PTSD, *r(18)*=.55, *p*=.012. No effect of sibling's age or closeness in age to sibling. Time since Index Child's death negatively associated with siblings' self-reports of PTSD, *r(18)=-.6, p*=.005.	6 (50%)
2a.Balk (1983a) 2b.Balk (1983b)	N=33, 20f (61%) and 13m (39%)). Recruited from self-help groups. Age at time of study: 14-19yrs. Cause of death: accident = 20 (61%) illness = 10 (30%); murder/suicide = 3 (9%). Time since death: 4-84	Observational Comparison with normative data	Age- 14-16 vs 17-19yrs Time since death	Self-Concept perceptions - **OSIQ**	No difference in siblings' self-concept from norms.	4 (33%)

	months (*M* =23.6; *SD*=16.4).					
3. Birenbaum et al. (1989)	N=61 (gender unreported). Recruited from hospital records (ages unreported). Cause of death: cancer. 4 time points: admission to study, 2 weeks after death, 4 months after death, 1 yr following death.	Longitudinal Comparison with normative data	Death of Index Child	Emotional and behavioural difficulties - **CBCL** parent and teacher report	Siblings had higher levels of internalising and externalising behaviour problems and lower levels of social competence than normative data (effect sizes unreported).	6 (50%)
4. Bolton et al. (2016)	N=7243, 3627f (50%), 3616m (50%). Recruited from cohort study of 1.2 million residents of Manitoba in Canada from 1984-2009. Age at time of death 0-39yrs. *M* = 9.3 *SD* = 7.3. 95% below age of 24[b]. Causes of death: accident =712 (22.4%) illness =1406 (44%) suicide, homicide, SIDs and perinatal =1067 (34%). Time since death: 2 yrs.	Longitudinal cohort Comparison group matched for demographics	Change 2 yrs pre-death and 2 yrs post-death Siblings older than 13 yrs at time of death vs siblings bereaved under 13 yrs	Depression, anxiety disorders, alcohol and drug use, ADHD and suicide attempts Physician diagnosed disorders	Bereaved siblings had greater rate of change in physician diagnosed mental health disorders compared to controls: 4.9% to 8% after death vs 4% to 5.3%. Bereaved siblings had greater rates of change in depression (ARR=2.71, 95% CI 1.94-3.79), *p*<.0001. Bereaved siblings had higher drug use, but rate of change was not significant, reflecting higher drug use pre-bereavement. Under 13s at time of death had greater rates of change for depression, ADHD, any mental health disorder and hospitalisations than over 13s. Under-13s had 7-fold increased rate in depression from pre-death to post-death. Adjusted relative rate (ARR) =7.25, 95%CI: 3.65-14.43 significantly greater than the rate in the 13+ age group (ARR) =2.27, 95% CI: 1.89-2.73.	11 (92%)

5a. Davies (1988)	N=55, 34f (62%), 21m (38%). Primary analysis one child, per family (21f (62%), 13m (38%)). Recruited from self-help groups. Age at time of study 6-19 yrs. Cause of death: cancer. Time since death: 2-6 months	Observational Comparison with normative data	Closeness index	Emotional and behavioural difficulties (parent-report) – **CBCL**	Bereaved children had higher scores for internalising behaviour and total scores compared to normed scores. No effect for closeness in age. Initial analysis revealed no association between closeness index scores and higher internalising and/or externalising scores. Secondary analysis comparing siblings with highest internalising scores (n=11) with those with lowest internalising scores (n=12) revealed those with highest internalising scores had highest scores on the closeness index.	4 (33%)
5b. Worden et al. (1999)	N=75, 41f (55%), 34m (45%) (including data from Davies (1988) above and McCown and Pratt (1985) below. Recruited from self-help groups. Age at time of study 6-18 yrs. Cause of death: mixed, majority illness. Time since death 2-13 months, *M* =6.92, *SD*= .62	Secondary analysis of data Cross-sectional Children bereaved by parental death, 65m (52%), 60f (48%) Recruited from self-help groups. Age at time of study 6-17. Cause of death mixed, majority illness. Time since death 2-13 months	Gender Age Months from death	Emotional and behavioural difficulties (parent-report) – **CBCL**	24% for parent loss group and 25 % for sibling loss group had emotional and behavioural problems. No differences in total number of problems nor in any of syndrome scales. Sibling bereaved girls were more anxious, depressed and had more thought problems than parentally bereaved girls. Effect sizes unreported. Siblings in same families had similar problems.	6 (50%)

6. Demi and Gilbert (1987)	N=18, 11f (61%), 7m (39%). Recruited from professional contacts. Age at time of study, 10 to 21.9 yrs, *M*= 14.7 yrs. Causes of death: accident = 5 (56%) illness = 3 (33%) suicide = 1(11%). Time since death: 4-24 months, *M* = 9.	Cross-sectional	Parents' emotional distress – **HSCL** Parents' grief reactions **– IES** Parental functioning – **PRS**	Emotional and behavioural difficulties (self report and parent-report) – **CDI and CBCL** Trauma – **IES**	Parents' emotional distress and siblings' emotional distress positively associated r=.47, p=.05. Parents' grief avoidance scores not associated with siblings' grief avoidance scores or intrusion scores. No association between parental role dysfunction and siblings' emotional distress. Parental role dysfunction and children's scores on the CBCL were positively associated, r=.52, *p*<.05.	3 (25%)
7. The Swedish Study. 7a. Eilegard et al. (2013b)	N=174, 101f (58%), 73m (42%). Recruited from hospital records. Aged 12-25 yrs at time of death. Mean age at time of study: 23 yrs. Cause of death: cancer. Time since death: 2-9 yrs *M*= 6.3 *SD=2.3.*	Cross-sectional Comparison group matched for demographics	Index Child's death	Anxiety and depression (self-report) – **HADS**	No difference in anxiety or depression between bereaved and non-bereaved siblings.	10 (83%)
7b. Wallin et al. (2015)[b]	See above.	Retrospective observational No comparison group	Perception of communication during illness after death	Anxiety and depression – (self report) **HADS**	Siblings reporting wanting to talk more about their feelings in the Index Child's last year of life had higher anxiety (15/58, 26%) than those satisfied with the amount they talked (13/115, 11%), relative risk (RR) =2.3 (1.2-4.5). Siblings reporting inability to talk to their family post-bereavement and avoided healthcare professionals in the hospital setting had higher risks of anxiety than those who could. Respectively (RR=2.5(1.3-4.8)) and (RR=6.7(2.5-18.2)).	4 (33%)

7c. Eilertsen et al. (2013)[b]	See above.	Retrospective observational No comparison	Perception of social support prior to and following death	Anxiety and depression (self report)- **HADS**	Siblings reporting unsatisfied need for social support had higher levels of anxiety than those who were satisfied at three points: during the Index Child's last month before death (RR=3.6(1.8-7.3)); after death (RR=2.9(1.5-5.6)) and at the time of the study (RR=3.8(2.0-7.2)). Siblings had higher anxiety who perceived that their parents (RR=2.7(1.3-5.5)) or neighbours (RR=5.4(1.3-21.9)) did not care for them at the time of death compared to those who perceived they were cared for.	4 (33%)
7d. Sveen et al. (2014)[b]	See above.	Retrospective observational No comparison	Time since death, age at study, age at death of sibling, gender, living with parents, employment, education, children, loss of significant person pre or post lost.	Grief – adapted from the (self-report) **ICG.**	Majority (54%) reported that they had worked through grief "not at all" or only "to some extent" at the time of the investigation. Longer time since loss and having social support needs met associated with more completely worked through grief. Sibling's age at time of death, gender, living with children, having dependent children, being employed, level of education, studying or loss of another significant person not associated with worked through grief.	7 (58%)
7e. Lövgren et al. (2016)[b]	See above.	Retrospective observational No comparison	Influence of siblings' experiences of death.	Anxiety and depression – (self report) **HADS**	Siblings reporting no one talked with them about what to expect when Index Child was dying had higher anxiety 2–9 yrs after the loss.	4 (33%)
8. Fletcher, Mallick, Song,	1. The Wisconsin Longitudinal Study (WLS), a sample of 10,317	Longitudinal cohort	Gender	From WLS – education, marriage divorce,	WLS sample: Bereaved females had reduced schooling compared to controls (.25 yr less).	10 (83%)

Wolfe (2013)	people graduating from Wisconsin in 1957. Outcomes for offspring who experienced death of a sibling before they were aged 25 yrs. N=850, 446f (52%), 482m (57%). Mean age at time of survey – 38 yrs (*SD* =4.7) (range unreported). 2. National Study of Adolescent Health (Adhealth), nationally representative sample of students in grades 7-12 in 1994-1995. Adhealth sample-N=771 470f (61%), 301m (39%). Mean age at end of study 29 yrs (*SD=1.75)* (range unreported) Causes of death unreported.	In WCLS Comparison with 17,342 total sample In Adhealth compared with 10,049 total sample		employment and co-residency. From Adhealth – education attainment, occupational success, marital history and residential independence.	Adhealth sample: Females and males had reduced schooling compared to controls (respectively approx. six months less and 3 months less). No effect for gender. Bereaved females had increased high school drop-out, decreased college attendance, reduced earnings, dependence on social programmes and increased rates of teen pregnancy. Bereaved males had lower test scores. Females more likely to be dependent on social programmes than males and those who had not experienced sibling bereavement. No other gender differences.	
9. Gerhardt et al. (2012)	N=105, 60f (57%), 45m (43%) Recruited from hospital records. Aged 8-17 at time of recruitment (*M* = 12.79 yrs (*SD*=2.56).	Cross-sectional Comparison group: classmates matched for	Gender, grade level and time since death.	Social relationships – (teacher , peers and self report) **RCP; PAR; BFN**	Bereaved siblings scored higher than on aggressive/disruptive behaviour, $F(1,102) = 2.77$ $p<.05$. Bereaved siblings were more prosocial by teacher report $F(1, 99) = 5.40$, $p < .05$, (d = .26).	10 (83%)

	Cause of death: cancer. Time since death: 3 – 19 months (*M* = 9.78, *SD* = 3.34).	gender, race and age			As whole, no differences between bereaved siblings and comparison classmates for peer acceptance ratings or total best friend nominations. Bereaved males, but not females perceived as more sensitive/isolated *F*(1,105) =6.94, *p*<.01) and more victimised, *F*(1,99) =5.48 *p<.01, d*=.45-.5. Bereaved siblings in elementary school had lower peer acceptance *F*(1,99) =5.02 *p=.05* and fewer best friend nominations *F*(1,103) =*3.91 p<.05*, *d=-.43 to -.4.* Bereaved siblings in middle/high school perceived by peers and teachers as higher on leadership popularity *F*(1,111) =5.79 *p*<.05 and *F*(1,104) =5.61 *p=.05, d*=0.28-0.41. No effects for time. No effects for self-report of social behaviour.	
10. Hogan (1988)	N=40, 25f (63%), 15m (37%). Recruited from self-help groups. Age at time of death: 13-18 yrs (*M*= 15.2). Cause of death: 63% sudden death, 37% expected. Time since death: 3-36 months	Cross-sectional Internal comparator divided into time since death: 3-18 months and 19-36 months.		Grief –(self report) **HSIB**	16 HSIB items correlated with 3-18 month period. 5 HSIB items correlated with 19-36 month period.	4 (33%)
11. Hogan and	N=127, 89f (70%), (30%) Recruited from self-help	Observational	Time after death:	Grief (self report)- **HSIB**	Positive association (high score = reduced symptom) between elapsed time grief score.	4 (33%)

Greenfield (1991)	groups. Aged 13-18yrs (mean age unreported). Cause of death: 73% sudden; 27% expected. Time since death: 33-84 months (*M*=30.7).	Internal comparator divided into time since death: 3-18 months and time since death:19-36 months.	3-18 months and 19-84 months. Self-concept scores association with grief scores- **OFSIQ**		1-18 months after death, siblings had mean score half an SD below normative scores on self-concept. After 18 months those with low levels of grief had high levels of self-concept; those with mean levels of grief had mean levels of self-concept and those with high levels of grief scored lower on self-concept. *F*(22,146) =3.1, *p*<.01.	
12. Martinson et al. (1987)	N=29, 19f (66%), 10m (34%). Recruited from families whose child had died in a homecare programme not hospital. Age at time of study 9-18 yrs, *M*= 14.17). Cause of death: cancer. Time since death: 7-9 yrs (*M* = 96.6 months)	Observational Comparison with normative data	Gender, time since death and age of sibling	Self-concept – (self-report) **P-H SCS**	Bereaved siblings scoring higher in self-concept than normative scores (high score = stronger sense of self-concept). No effect for gender, time since death and age of sibling.	4 (33%)
13. McCown and Pratt (1985)	N=65 (32f (49%), 33m 51%)) from 44 families. Recruited from hospital and self help groups. Age at time of study 4-16 yrs (*M*= 9.5 yrs). Causes of death: unreported.Time since death: 2-13 months.	Observational Comparison with normative data	Age, gender, length of illness, family size, funeral attendance, maternal status, time since death, before death behaviour, parent child-communication.	Emotional and behavioural difficulties (mother report) – **CBCL**	Bereaved siblings had higher behaviour problems compared with CBCL norms, *t(*65) =- 3.15 <0.01. Small negative correlation between number of children in family and the child's behaviour problems. Attendance at funeral associated with behaviour problems, *t*(65) =3.29, *p*<.01. Death in hospital rather than home associated with behaviour problems, *t*(65) =2.1, *p<.05.*	5 (42%)

					Age, sex, length of illness and parental communication or their weighted combination associated with CBCL scores. No effect on CBCL scores for marital and employment status of mothers and time since death.	
14. Morris et al. (2016)	N=62, 40f (65%), 22m (35%). Recruited from hospital records. Age at time of study 8-18 yrs, *(M*= 12.95, *SD= 3.59).* Cause of death: accident = 4 (6%) medical illness = 51 (82%) other = 6 (12%). Time since death: 2.91 yrs (*SD* 1.61) – range .5-5 yrs.	Cross-sectional Comparison group: 60 mothers 28 fathers	Caregiver PTSD PGD and depression symptoms - (PDSD-Ri, DASS, PG-13) and sibling report of parenting behaviours (APQ) .	Trauma (self-report) **PTSD Ri, PG-13, PTSD-CL, CDI**	Maternal and siblings' symptoms of PTSD (*r* =.55, *p*<.01), depression (*r*=.43, *p*<.01), and PGD (*r* =.32, *p*<.05) positively associated. Paternal depression symptoms positively associated with siblings' depression (*r* =.67, *p*<.01). Significant effect of gender with girls being more affected by paternal symptoms than boys *(r= -.65,* p<.05). Paternal symptoms, but not maternal symptoms, were associated with less positive parenting behaviours (depression *r*=-.62; PTSD, *r*=-.59) and PGD, *r*=-.41). Lower parental involvement in association with paternal (not maternal) symptoms (depression *r*=-.54; PTSD, *r*=-055) and PGD, *r*=-.42). Parental involvement not a mediating factor between parental symptoms and sibling symptoms when gender and age included as co-variates.	7 (58%)
15. Mulhern et al. (1983)	N=45, 30f (67%), 15m (33%). Recruited from hospital records. Age at time of study 4-13 yrs, (*M* = 7).	Cross-sectional Comparison group: siblings where Index Child died not	Parental psychopathology (**MMPI-138)**	Emotional and behavioural difficulties **LBC**	Siblings of children who died in Homecare programme had fewer total emotional difficulties. Where the Index Child had not died in the Homecare programme, siblings displayed fear and neurotic behaviour above criterion for clinical significance.	5 (42%)

	Time since death: 3-29 months (*Mdn* 13-14). Cause of death: cancer.	in a Homecare programme vs siblings where Index Child died within programme.	Family functioning **(FES)**			
16. Pettle Michael and Lansdown (1986)	N=28 (gender unreported) (ages 5-21yrs). Recruited from hospital records. Time since death: 18-30 months. Cause of death: unreported.	Cross sectional Comparison with normative data	Self-concept and behaviour.	Self-concept – **SCS** Emotional and behavioural difficulties - **RCS** (parent and teacyher report)	43.5% of the children scored above clinical cut-offs for emotional and/or behavioural problems vs 6.8% for children aged 10-11 yrs in general population, 20.8% for boys and 13.6% for girls aged 13-14 yrs. Children perceived themselves to be different from their ideal self and their dead sibling t=1.84 p<.05. Children participating in fewer events around death had lower self-esteem.	3 (25%)
17. Rosenberg et al. (2015)	N=58, 40f (69%), 18m (31%). Age at time of study: Mean = 25.6 yrs *SD* 7.8. Recruited from previous research study. Cause of death: cancer. Time since death: *M* = 11.8 years *SD*= 3.2.	Observational Comparison with normative data	Length of time since Index Child's death. Social support Perceptions of the illness experience.	Anxiety and depression - **Kessler-6**. Social support - **MOS- Social support**	Anxiety and depression for bereaved siblings in the normal range. Siblings reporting peer relationships negatively impacted by Index Child's cancer had higher distress scores, β=4.3 CI (1.8,6.8), and lower social support scores, β=-45.4 (CI -29.5, -61.3). Siblings dissatisfied with information from their parents had higher distress scores, β=3.4(CI 1.1 -5.7) and lower social support scores, β=-45.7(CI 31.3-60.1). Siblings dissatisfied with information from health care staff when their sibling was dying or who did not feel prepared for their sibling's death had higher distress,	5 (42%)

					β=2.6(CI 0.7,4.5) and lower social support β=-22.8(CI -2.1-43.5). Siblings had higher distress scores, but not lower social support scores who did not say goodbye β=2.2(CI 0.3,4.0) and did not believe they had worked through grief compared to those who believed they had β= 3.4 (CI 1.9, 49).	
18. Stikkelbroek et al. (2016)	N=15 (gender unreported) Mean age at T1 of study = 11.14 yrs *SD* 0.53) Mean age at T4 of study = 19.24 yrs *SD* = 0.65*)*. Cause of death: unreported. Time since bereavement: within 2 yrs.	Longitudinal cohort Comparison group: 55 parentally bereaved adolescents in same sample.	Pre-loss mental health problems.	Emotional and behavioural difficulties (reported by siblings) **YSR, ASR**	Bereaved participants (comprising 55 parentally bereaved and 15 siblings) had significantly increased internalising problems compared to the non-bereaved *t*(1168) = -3.97, *p*<.001 (Cohen's *d* pooled .37; 95% CI .13-.62). No difference between sibling bereaved adolescents and parentally bereaved adolescents in internalising problems.	8 (67%)

[a]The original intention in reviewing this study was to report the data in relation to siblings bereaved from accident. However, because no effect was found in relation to cause of death the authors did not report the data from siblings bereaved through accident and homicide separately and so these results report on psychosocial outcomes where 50% of Index Children who died through homicide.

[b]Although this study included siblings aged over 25, over 95% of siblings were under the age of 25 at the time of the Index Child's death it was decided to include the data.

Narrative Synthesis of Results

The qualitative narrative synthesis is organised into three sections: (1) short-term psychosocial outcomes (i.e.within two years of death of the Index Child) in light of previous evidence that most bereaved children recover within this period (Currier et al., 2007; Dowdney, 2000; Worden & Silverman, 1996) (see Table 1.3); (2) long-term psychosocial outcomes (see Table 1.4); and (3) factors associated with short-term and long-term outcomes (see Table 1.5).

Short-term Outcomes

No studies with validated outcome measures investigated the outcomes of bereavement within the first two months of death of the Index Child and so this review does not consider symptoms of acute grief. Seven studies reported the psychosocial outcomes of bereavement within 2-24 months of the Index Child's death using either a comparison group or comparison with normative data (Balk, 1983a, 1983b; Birenbaum et al., 1989; Bolton et al., 2016; Davies, 1988; Gerhardt et al., 2012; McCown & Pratt, 1985; Pettle Michael & Lansdown, 1986).

Mental health outcomes. Five studies investigated the relationship between sibling bereavement and emotional stress or mental health problems. In a cohort study, Bolton et al. (2016) considered prevalence of mental health disorders two years prior to an Index Child's death compared with two years after death with a matched control group of siblings. Bereaved siblings had a significantly greater increase in physician-diagnosed mental health disorders compared to control siblings (i.e., 4.9% to 8% after death vs control 4% to 5.3%). In terms of specific outcomes, increased rates of depression and mental health related hospitalisations were demonstrated. This robust study (quality rating 92%) indicates that, even taking into account the higher rates of mental health problems before the sibling died, there were significant mental health consequences for bereaved siblings.

Four studies examined siblings' emotions and behaviour using measures with normative data. Three used the Children's Behaviour Checklist (CBCL; Achenbach & Edelbrock, 1983) to assess children between two and 13 months after the Index Child had

died (Birenbaum et al., 1989; Davies, 1988; McCown & Pratt, 1985). Birenbaum et al. used both teacher and parent-report measures and did not report effect sizes. McCown and Pratt (1985) and Davies (1988) used only parent-report measures and reported increase rates in total problems on the CBCL scale with effect size of r= .36 and r=.27 respectively. Pettle Michael and Lansdown (1986) used a different parental and teacher report scale (the Rutter Child Scale (RCS)) and also found significantly higher rates of difficulties (effect sizes unreported). Studies consistently indicate an association with mental health problems in comparison with the wider population in the first two years after sibling bereavement, but relied on parent-report, had small sample sizes and were of poor quality as reflected in quality ratings ranging from 25-50%.

Social outcomes. One study investigated short-term social outcomes for bereaved siblings, finding no difference in social relationships between bereaved siblings compared to their peers. Gerhardt et al. (2012) compared behaviour and peer acceptance scores of siblings 3-19 months after the death of the Index Child from cancer with matched peers. The Revised Class Play (RCP; Masten et al., 1985) assessed behaviour and Peer Acceptance Ratings (PAR; Asher, Singleton, Tinsley, & Hymel, 1979) and Best Friends Nominations (BFN; Bukowski & Hoza, 1989) were assessed friendships. Group comparisons found little evidence of group differences for any of the RCP domains of leadership-popularity, prosocial, aggressive-disruptive, sensitive-isolated and victimisation, or for PAR and social preference scores. This study, with a quality rating of 83%, was cross-sectional and only considered siblings bereaved through cancer, but the use of matched controls and third party measures means that it has considerable value.

Self-concept. One study explored the effect of bereavement on adolescents' self-concept in the short to medium time between 4-84 months after the death of their sibling (Balk, 1983a, 1983) using the self-image questionnaire (OFSIQ; Offer et al., 1977). Although for some adolescents in the group, lapse of time since the death was more than 24 months, the median period was 22.7 months and so findings are included within this short-term outcome section. Self-concept was measured on 11 different dimensions: impulse control, emotional tone, body and self image, social relationships, moral values, sexual attitudes, family relationships, mastery of external world, vocational educational goals, psychopathology and ego strength. Bereaved adolescents did not have significantly different self-concept scores relative to normative data. This result appears to conflict with

the studies above but there is a high risk of bias in this study with all families being recruited from self-help groups and most being of higher socio-economic status (quality rating 33%). The result may also reflect that the median time from the death of the Index Child was almost two years or that self-concept is not a construct which changes as a result of bereavement.

Long-term Outcomes

Four studies investigated long-term psychosocial outcomes using a comparison group or a measure with normative data: a cohort study, a cross-sectional study and two observational studies (Eilegard et al., 2013b; Fletcher et al., 2013; Martinson et al., 1987; Rosenberg et al., 2015).

Mental health outcomes. Two studies investigated long-term mental health outcomes of a sibling dying of cancer finding no significant difference in anxiety or depression in the bereaved sample compared to controls or normative data. Eilegard et al. (2013b) investigated psychological outcomes for 174 siblings bereaved through cancer 2-9 years previously compared to 219 siblings matched according to age, gender and region (the Swedish Study). They found no significant difference between groups in levels of anxiety and depression measured using the self-report Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983). Given the use of matched controls, this is a relatively robust finding as reflected in the 83% quality rating.

Rosenberg et al. (2015) also investigated long-term effects for siblings bereaved through cancer (mean time since the death of the Index Child had died was 11.8 years) in a study using normative data instead of a control group. The lack of clear hypotheses and control group resulted in a lower quality rating of 42%. Participants completed two self-report measures with normative data for the US population: the Kessler-6 Psychological Distress Scale (Kessler et al., 2003) a screening measure of anxiety and depression and a measure of social support, the MOS (Sherbourne & Stewart, 1991). On both measures, participants' scores were not significantly different from the population. Although these two studies are focused on a specific group of bereaved siblings (i.e. those bereaved through cancer), they are from different countries and provide consistent evidence that in the long-term psychological distress amongst bereaved siblings levels returns to normal.

Social outcomes. Fletcher et al. (2013) investigated the effects of sibling bereavement before the age of 25 on adult outcomes of educational attainment, occupational success and marital history. They examined data from two USA cohort studies: the Wisconsin Longitudinal Study (WLS), a sample of 10,317 women and men who graduated from Wisconsin in 1957; and the National Study of Adolescent Health (Adhealth), a nationally representative sample of students in grades 7-12 in 1994-1995. In relation to the WLS cohort, outcomes for 850 of their offspring who experienced death of a sibling before they were 25 years were compared with 17,342 offspring with only healthy siblings (4.7% of total). In Adhealth, 771 individuals who had experienced the death of a sibling before they were 25 were compared with 10,049 who had not (7.1% of total).

Amongst the WLS sample, siblings who were bereaved before they were 25 had reduced schooling. Results of the Adhealth study are more difficult to interpret because they are reported by gender (see below), but they indicate that women (not men) had increased high school drop-out, decreased probability of college attendance, reduced earnings rate, dependence on social programmes and increased teen pregnancy rates. In contrast, bereaved boys only had significantly lower test scores. This good quality study with a quality rating of 83% indicates that death of a sibling has long-term effects. Outcomes such as reduced earning and dependence on social programmes may be long-term sequelae of shorter term distress which resulted in increased rates of pregnancy or reduced schooling.

Self-concept. One study explored long-term effects of sibling bereavement on self-concept (Martinson et al., 1987). This study explored association of sibling bereavement with self-concept of 29 children aged 8-18, seven to nine years after death of the Index Child by comparing to normative data on the Piers-Harris Self-Concept Scale (PHSCS; Piers, 1969). Contrary to expectations, bereaved siblings had signficantly higher self-concept scores relative to normative scores (effect sizes unreported). However, methodological problems with this study include a lack of clear hypotheses and a wide age range (quality rating 33%). It also seems likely that self-concept will have considerable variation within this age group. Furthermore, all Index Children died in a home care programme and this means results are likely not to be generalisable to other bereaved populations as there is some evidence that children supported on the home care

programme may have fewer problems resulting from sibling bereavement (see Mulhern, Lauer and Hoffman (1983) below).

Factors Associated with Psychosocial Outcomes

A key factor associated with psychosocial outcomes was lapse of time. This is considered first. Other factors associated with psychosocial outcomes are organised by Stroebe et al.'s (2006) integrative risk factor framework.

Lapse of time. Six studies reported association between time since the death of the Index Child and level of distress (Applebaum & Burns, 1991; Birenbaum et al., 1989; Gerhardt et al., 2012; Hogan, 1988; Hogan & Greenfield, 1991; Sveen et al., 2014). Two studies found no effect for time: Birenbaum et al. (1989) found no association of time in levels of emotional or behavioural distress measured on the CBCL within the first year after bereavement and Gerhardt et al. (2012) also found no association between time and social relationship in a study of siblings 3-19 months ($M = 9.78$, $SD = 3.34$) after death of the Index Child. Although these two studies were focused on different psychosocial outcomes and Gerhardt et al.'s study was more robust (quality rating 83%) than Birenbaum et al.'s study (quality rating 50%), they both focused on outcomes in the relatively short term.

Three studies focused on a longer period since death found that poor psychosocial outcomes were associated with less time elapsed since the death. In a study of adolescents' grief symptoms 3-36 months after the death of the Index Child (measured by HSIB (Hogan, 1990)), Hogan (1988) found no effect of time when investigating differences between the first 12 months and the second 12 months after death, but those whose sibling had died over 18 months previously had significantly reduced grief scores (no effect size reported). Hogan and Greenfield (1991) similarly found that grief in adolescents measured by the HSIB (3-84 months after death of Index Child) was negatively associated with time since death.

Applebaum and Burns (1991) found that trauma symptoms measured by the Child Reaction Index (CRI; Frederick, 1985) in siblings bereaved between 4-77 months (median 35) previously were negatively associated with amount of time passing. Also consistent with these findings, in a paper reporting on data from the Swedish Study, Sveen et al.

(2014) found that more time since death was associated with more fully worked-through grief. There are methodological problems with these studies: Hogan and Hogan and Greenfield (both quality rating 33%) recruited from self-help groups and did not report hypotheses clearly; Applebaum and Burns' study (quality rating 50%) included a wide age range and 50% of the Index Children died as a result of homicide; and Sveen et al (quality rating 58%) relied on retrospective reporting. However, the results provide a consistent picture that grief and distress reduce over time, but possibly not significantly within the first year.

Loss-oriented stressors. Four studies reported findings related to loss-oriented stressors (e.g. relationship with the person who had died and quality of that relationship) (Applebaum & Burns, 1991; Davies, 1988; Stikkelbroek et al., 2016; Worden et al., 1999). All studies were focused on stressors associated with short-term outcomes.

Sibling bereavement compared with parental bereavement. Two studies compared psychosocial outcome between sibling bereavement and parental bereavement. Stikkelbroek et al. (2016) found no significant difference when they compared internalising symptoms of sibling bereaved adolescents to parentally bereaved adolescents using the Youth Self-report (YSR; Achenbach, 1991) within two years of death. However, although this was a longitudinal cohort study with a medium quality rating of 67%, the focus of the study was primarily parental bereavement and the sibling sample size was small (n=15) so there may have been insufficient power to find differences in effect. In a medium quality study (quality rating 50%), Worden et al. (1999) conducted a secondary analysis on data from three studies, with 200 participants in total (75 sibling bereaved and 125 parentally bereaved) (see Davies, 1988; McCown & Pratt, 1985; Worden & Silverman, 1996) and compared sibling and parental bereavement outcomes 2-13 months after death using the CBCL finding no significant difference between the two groups. Although the evidence is limited, these studies indicate that the death of a sibling is an important loss with similar psychosocial outcomes to parental bereavement in the short-term.

Closeness of sibling. Two studies investigated whether closeness of a sibling to the Index Child prior to their death was associated with greater difficulties (Applebaum & Burns, 1991; Davies, 1988). Applebaum and Burns (quality rating 50%) found no

association of closeness of age of sibling and Index Child and traumatic distress. However, since the cause of death of 50% of the Index Children in this study was homicide, it is possible that levels of traumatic distress were particularly high which could have masked an effect of closeness. Davies (1988) also found no significant association between closeness index and emotional and behavioural difficulties on the parent-reported CBCL although a secondary analysis in which those with the highest internalising scores (n=11) were compared with those with lowest internalising scores (n=12) indicated that those in the first group had the highest scores on the closeness index. A bias issue in Davies' study (quality rating 33%) is the use of a parent-report measure of distress and that closeness was also reported retrospectively by parents. The high risk of bias in these studies means that findings should be treated cautiously.

Restoration oriented stressors. Four studies investigated restoration-oriented stressors (e.g. consequences from circumstances of death) (Eilertsen et al., 2013; McCown & Pratt, 1985; Mulhern et al., 1983; Pettle Michael & Lansdown, 1986; Rosenberg et al., 2015). Three studies investigated short-term consequences: Mulhern et al. (1983) (quality rating 42%) investigated differences between siblings where the Index Child had died from cancer on a homecare programme with those where the Index Child had not been on the homecare programme using the Louisville Behaviour Checklist (LBC; Miller, 1984), a parent-reported measure of emotional and behavioural difficulties. There was a significant difference between the two groups with the homecare group of siblings presenting behaviours within normal limits whereas siblings of patients who were not provided homecare displayed fear and neurotic behaviour in which the mean standard scores were above the criterion for clinical significance. However, different profiles between the two groups (families providing homecare displaying greater family commitment/support) suggest that the differences between the groups' level of distress may have been explained by differences in family dynamics more generally. Pettle Michael and Lansdown (1986) (quality rating 25%) found that children who were involved in more events around the Index Child's death (e.g. knowing about the child's diagnosis and illness or having the sibling die at home) had higher levels of self-esteem. These two studies' results are consistent, although they both have a high risk of bias. In contrast to the two studies above, McCown and Pratt (1985) (quality rating 42%) found increased behavioural difficulties in bereaved children who attended the funeral. However, the study provides

no further detail of the children's involvement and this finding was not underpinned by a hypothesis.

Two studies explored the long-term effects of events at the death. Secondary analysis of data from the Swedish study using the same sample of 174 participants explored the association between anxiety and siblings' retrospective reports relating to communication in the last year of the Index Child's illness and death (Eilertsen et al., 2013; Lövgren et al., 2016; Wallin et al., 2015). The quality rating of 33% for each paper was lower than for the original paper by Eilegard et al.'s (2013b) because of reliance on retrospective reports and lack of comparison groups. Compared to those who did not report such problems, individuals were at higher risk of anxiety if they reported the following: that they would have liked to talk more about their feelings in the Index Child's last year of life *RR*=2.3 (1.2-4.5); that they could not talk to their family after the bereavement *RR*=2.5(1.3-4.8); and they avoided healthcare professionals for fear of being in the way in the hospital setting *RR*=6.7(2.5-18.2) (Wallin et al., 2015). Siblings who reported that no-one talked with them about what to expect when their brother/sister was going to die also had higher levels of anxiety 2–9 years after the loss (Lövgren et al., 2016). Rosenberg et al. (2015) also found that participants who reported peer relationships were negatively impacted by sibling's death or were dissatisfied by the information they were given when their sibling was dying and did not feel prepared for their sibling's death, had higher distress scores and lower social support scores compared to those with those who did not report these experiences. This study also relied on retrospective reporting as reflected in a quality rating of 42%.

Although studies report consistent findings, the short-term studies rely on parental report of behaviour while both long-term studies rely on retrospective reporting and so there is a high risk of bias and direction of causation is unknown. Nonetheless, it seems likely that involvement in the sibling's death and being able to talk are important in increasing likelihood of better outcomes.

Intrapersonal Risk Factors. Seven studies reported findings relating to intrapersonal risk factors of age, gender or self-concept (Applebaum & Burns, 1991; Bolton et al., 2016; Fletcher et al., 2013; Gerhardt et al., 2012; Hogan & Greenfield, 1991; McCown & Pratt, 1985; Worden et al., 1999).

Age. Four studies reported findings in relation to age of the sibling at time of bereavement. Bolton et al. (2016) conducted analyses for disorder prevalence and risk factor by age at time of death (younger or older than 13 years). Siblings under 13 years had higher increased rates for depression (adjusted relative rate (ARR) 7.25 vs 2.27), ADHD (ARR 1.69 vs 0.47), any mental disorder (ARR 2.06 vs 1.50) and physician visits for mental illness (ARR 3.06 vs 1.66) than those over 13. The over 13s were particularly at risk of drug use. Gerhardt et al. (2012) investigated social relationships and found that bereaved siblings in elementary school, but not in middle/high school, were perceived by their peers as less prosocial, and more sensitive/isolated (d=-.38-.65) and had significantly lower peer-acceptance and fewer best-friend nominations relative to classmates. Bereaved siblings in middle/high school grades were perceived by peers and teachers as significantly higher on leadership popularity although these effects were small (d=.28-.41). Although two studies found no association between age and psychosocial outcome (Applebaum & Burns, 1991; McCown & Pratt, 1985), their sample sizes were small and their primary focus was not age (quality rating 50% and 42%). Despite mixed results, the two most robust studies (with quality ratings of 92% and 83% respectively) provide consistent evidence that in the short-term younger children appear to have worse psychosocial outcomes than older children (i.e those over eleven years).

Gender. Three studies focused on how gender influences psychosocial outcomes of bereavement (Fletcher et al., 2013; Gerhardt et al., 2012; Worden et al., 1999). Two focused on short-term outcomes. In their study of the effect of bereavement on social relationships using matched classmates as a comparison group (quality rating 83%), Gerhardt et al. (2012) found that bereaved boys, but not girls, were perceived by teachers as more sensitive isolated and victimised (d=.45-.5) but there were null findings in all other domains. Worden et al. (1999) (quality rating 50%) found that girls who lost a sibling experienced signficantly more anxiety, depression and thought problems than girls who had lost a parent and parentally-bereaved boys were more likely to score highly on anxious behaviour (effect sizes unreported). These two studies appear to provide conflicting evidence about the gender effects of sibling bereavement, but were measuring different outcomes. Furthermore, the use of fourteen separate analyses in Worden's study calls into question the value of its result because of the likelihood of a Type I error.

Fletcher et al. (2013) focused on the long-term association of gender with psychosocial outcomes in a cohort study with a quality rating of 83%. In the WLS sample there were no gender effects. Amongst the Adhealth sample, measures of occupational attainment and social measures were compared and results reported by gender. There was limited evidence of significant differences in outcome by gender, although bereaved sisters were significantly more likely to be dependent on social welfare than bereaved brothers. Nonetheless, a wider range of significant differences relative to controls were reported for bereaved females, including increased high school drop out, decreased probability of college attendance, reduced earnings and increased rates of teen pregnancy. In contrast, bereaved males only had significantly lower test scores than controls. Thus there was some evidence that sibling bereavement affects girls more than boys in the long-term.

Self-concept. One study considered how a sense of self-concept was associated with grief in adolescents. Hogan and Greenfield (1991) found that those with low levels of grief after 18 months (measured with the HSIB) had high levels of self-concept indicating a more integrated sense of self (measured with the OSIQ); those with medium levels had mean levels of self-concept; but those with the highest grief scores had the lowest self-concept scores. Although this study indicated an association between self-concept and grief, the direction of causality is unknown and there was a high risk of bias with a quality rating of 33%.

Interpersonal factors. Seven studies reported on interpersonal factors. Two reported on the effects of social support. Five studies reported on how a sibling's distress is associated with the distress of other family members.

Social support. Two reported on the long-term effects of social support at time of death (Eilertsen et al., 2013; Rosenberg et al., 2015). Data from the Swedish Study indicated that dissatisfaction with social support compared to those who were satisfied was also associated with a higher level of anxiety (Eilertsen et al., 2013). Risk of anxiety was increased for those dissatisfied with support during the Index Child's last month before death, *RR*=3.6(1.8-7.3), after death, *RR*=2.9(1.5-5.6), and at the time of the study, RR=3.8(2.0-7.2). Risk of anxiety also increased for those who perceived that their parents or neighbours did not care for them at time of death, respectively *RR*=2.7(1.3-5.5) and *RR*=5.4(1.3-21.9). Rosenberg et al. (2013) also found an association between perceptions of lower levels of social support and greater distress, both retrospectively at the time of the

child's death and at the time of the study. Note though that, as above in relation to communication, participants were giving retrospective accounts of social support and so there is a high risk of bias.

Association with other siblings' distress. Two studies reported positive associations between the levels of distress of siblings in the same family (effect sizes unreported) (Birenbaum et al., 1989; Worden et al., 1999). However, both studies used the parent-report CBCL as a measure for distress and so this may reflect parental, not sibling experience.

Association with parental distress. Three studies examined family dynamics and particularly the association between parental distress and sibling distress (Applebaum & Burns, 1991; Demi & Gilbert, 1987; Morris et al., 2016). In the most robust of these studies (quality rating 58%), Morris et al. (2016) explored associations between parental symptoms of post traumatic stress disorder (PTSD) measured by the PTSD Checklist (Weathers, Litz, Herman, Huska, & Keane, 1993) and prolonged grief disorder assessed by the Prolonged Grief Disorder Questionnaire (PG-13; Prigerson et al., 2009) and child symptoms of PTSD measured by the Child version of Los Angeles PTSD Reaction Index (PTSD-Ri; Rodriguez, Steinberg, & Pynoos, 1999) and depression measured by the Child Depression Inventory (CDI; Kovacs & Beck, 1977). They also investigated whether parenting behaviours mediated such associations. Significant associations were found between maternal and sibling's symptoms of PTSD (r=.54), depression (r=.40) and PGD (r=.55). Paternal depression symptoms were significantly related to siblings' depression (r=.56), but other paternal associations were not significant. There was some evidence that girls were more affected by paternal symptoms than boys. Paternal symptoms, but not maternal symptoms, were associated with less positive parenting behaviours (depression r=-.62; PTSD, r=-.59) and PGD, r=-.41). Parenting quality therefore had an indirect effect on offspring mental health for fathers only. Although this study is correlational and so cannot provide a direction of causation, it highlights the association of parental and child grief reactions.

Applebaum and Burns (1991) investigated the association between parental and siblings' PTSD reactions to death of the Index Child (measured respectively by the structured interview enquiring about the DSM-III-R criteria and by Child Reaction Index (CRI; Frederick, 1985)). Parental participants were almost exclusively mothers (90%).

There was a significant positive relationship between parents' self-reports of PTSD and siblings' self-reports of PTSD (r=.55). The quality rating for this study was 50% and results must be interpreted in light of the fact that 90% of families were recruited from support groups and that the cause of death of 50% children was homicide. However, the study provides further support that there is an association between maternal distress and sibling distress.

In a study of nine families, with 18 siblings and 14 parents participating (making up 22 parent-child pairs), Demi and Gilbert (1987) investigated the association between parents' emotional distress and functioning and siblings' behaviour and mood between 4-24 months after the death of the Index Child. Parental distress and PTSD was measured respectively by the Hopkins Symptom Checklist (Derogatis, Lipman, Rickels, Uhlenhuth, & Covi, 1974)) and the Impact of Event Scale (IES; Horowitz et al., 1979. Parental functioning was measured by the Parental Role Scale (Weissman & Bothwell, 1976). Siblings' mood and behaviour were measured using the CDI and the CBCL. They found significant positive relationship between parents' emotional distress and siblings' emotional distress (r=.47) and a positive relationship between parental role dysfunction and siblings' behaviour problems on the CBCL (r=.52). In contrast, patients' IES avoidance scores were not significantly associated with siblings' avoidance scores or intrusion scores and there was not a positive relationship between parental role dysfunction and siblings' emotional distress. Results of this small study should be treated with caution since data from the families were used several times as reflected in the quality rating of 25%. Despite the varying quality of these studies they consistently indicate that parental (especially maternal) distress is associated with child distress. They also indicate that some of the child's distress may be mediated by negative parenting (especially by fathers).

Table 1. 3

Summary of short-term psychosocial outcomes with quality rating.

Study	*Psychosocial outcome and measure*	*Sibling bereavement associated with poor outcome*	*Quality rating*
	Mental health outcomes		
Bolton et al. (2016)	Prevalence of mental health problems (clinical records)	Yes	11 (92%)
Birenbaum et al. (1989)	Emotional and behavioural difficulties (CBCL)	Yes	6 (50%)
McCown and Pratt (1985)	Emotional and behavioural difficulties (CBCL)	Yes	5 (42%)
Davies (1988)	Emotional and behavioural difficulties (CBCL)	Yes	4 (33%)
Pettle Michael and Lansdown (1986)	Emotional and behavioural difficulties (Rutter scale)	Yes	3 (25%)
	Social outcomes		
Gerhardt et al. (2012)	Social relationships (PAR, BFN)	No	10 (83%)
	Self-concept		
Balk (1983a); Balk (1983b)	Self-concept (OSIC)	No	4 (33%)

Table 1. 4

Summary of long-term psychosocial outcomes with quality rating.

Study	*Psychosocial outcome and measure*	*Sibling bereavement associated*	*Quality rating*
	Adult social measures		
Fletcher et al. (2013)	Marital status, employment,	No	10 (83%)
Fletcher et al. (2013)	Length of education	Yes – reduced schooling	10 (83%)
	Mental Health Problems		
Eilegard et al. (2013)	Anxiety and low mood (HADS)	No	10 (83%)
Rosenberg et al. (2015)	Anxiety and low mood (K-6)	No	5 (42%)
	Level of social support		
Rosenberg et al. (2015)	Social support (MOS)	No	5 (42%)
	Self-concept		
Martinson et al. (1987)	Self-concept	Yes – better self-concept	4 (33%)

Table 1. 5

Summary of factors associated with outcome with quality rating

Factor associated with outcome/comparison group	***Study***	***Psychosocial outcome***	***Association between factor and outcome***	***Quality rating***
Lapse of time				
Within year of death	Birenbaum et al. (1989)	Emotional and behavioural difficulties (CBCL)	No	6 (50%)
4 -77 months after death	Applebaum and Burns (1991)	PTSD	Yes	6 (50%)
3-36 months after death	Hogan (1988)	Grief	Yes	4 (33%)
2-84 months after death	Hogan and Greenfield (1991)	Grief	Yes	4 (33%)
2-9 years after death	Sveen et al. (2014	Grief	Yes	7 (58%)
Loss oriented stressors				
Compared to parentally bereaved children	Stikkelbroek et al. (2016)	Internalising symptoms	No (groups had no significant differences)	8 (67%)
Compared to parentally bereaved children	Worden et al. (1999)	Emotional and behavioural problems	No (groups had no significant differences)	6 (50%)
Closeness of relationship	Davies (1988)	Emotional and behavioural problems	Yes, but only on secondary analysis of data.	4 (33%)
Closeness in age of sibling	Applebaum and Burns (1991)	PTSD	No	6 (50%)
Restoration-oriented stressors- short term				
Index Child had hospital care vs home care	Mulhern et al. (1983)	Emotional and behavioural difficulties	Yes (more problems if in hospital)	5 (42%)
Involvement in events around sibling's death	Pettle Michael and Lansdown (1986)	Emotional and behavioural difficulties	Yes (fewer problems if more involved)	3 (25%)
Restoration-oriented stressors- long term				
Retrospective report of lack of communication with family members or	Rosenberg (2015)	Anxiety and depression	Yes (more problems if lack of communication	5 (42%)

Factor associated with outcome/comparison group	***Study***	***Psychosocial outcome***	***Association between factor and outcome***	***Quality rating***
health care professionals				
Retrospective report of lack of communication with family members or health care professionals	Wallin et al. (2015) Lovgren et al. (2016)	Anxiety	Yes (more problems if lack of communication)	4 (33%)
Interpersonal risk factors –age				
Under 13 years and over 13 years	Bolton et al. (2016)	Mental health problems	Yes	11 (92%)
Under 10 years and over ten years	Gerhardt (2012)	Social relationships	Yes	10 (83%)
Age range 3-23 years	Applebaum and Burns (1991)	PTSD	No	6 (50%)
Age range 4-16	McCown and Pratt (1985)	CBCL	No	5 (42%)
Interpersonal risk factors - gender				
Male/female	Gerhardt et al. (2012)	Leadership-popularity, prosocial, aggressive-disruptive and peer acceptance and	No	10 (83%)
Male	Gerhardt et al. (2012)	Victimisation and isolation	Yes	10 (83%)
Female	Worden et al. (1999)	Anxiety, depression and thought problems	Yes	6 (50%)
Female	Fletcher et al. (2013)	Social welfare	Yes	10 (83%)
Interpersonal risk factors – self concept				
Self-concept	Hogan and Greenfield (1991)	Grief	Yes (lower self-concept associated with higher grief)	4 (33%)
Intra-personal risk factors- social support				
Retrospective report of dissatisfaction with social support	Eilertsen (2013)	Anxiety	Yes (more problems if lack of social support)	4 (33%)

Factor associated with outcome/comparison group	*Study*	*Psychosocial outcome*	*Association between factor and outcome*	*Quality rating*
Retrospective report of dissatisfaction with social support	Rosenberg et al. (2015)	Current social support Anxiety and depression	Yes (more problems if lack of social support)	5 (42%)
Intra-personal risk factors - parental distress				
Maternal PTSD, depression and grief	Morris et al. (2016)	Child PTSD and depression	Yes	7 (58%)
Paternal depression		Child depression	Yes	
Paternal PTSD and grief		Child PTSD and depression	No	
Maternal symptoms		Parenting behaviour	No	
Paternal symptoms		Parenting behaviour	Yes	
Parental PTSD (90% women)	Applebaum and Burns (1991)	Child PTSD	Yes	6 (50%)
Parental distress	Demi and Gilbert (1987)	Child distress	Yes	3 (25%)
Parental role dysfunction		Child behaviour problems	Yes	
Parental role dysfunction		Child distress	No	
Parental avoidance of emotions		Child avoidance of emotions or intrusion	No	
Intrapersonal risk factors - sibling distress- short term				
Sibling's emotional and behavioural problems	Birenbaum et al. (1987)	Emotional and behavioural problems	Yes	6
Sibling's emotional and behavioural problems	Worden et al. (1999)	Emotional and behavioural problems	Yes	6

Discussion

Psychosocial Outcomes

This systematic review confirms that within two years of the death of their sibling, bereaved children have increased risks of mental health difficulties and emotional and behaviour problems compared with children who have not been bereaved. Bolton et al.'s (2016) finding that bereaved siblings had a higher risk of mental health problems even allowing for social-economic status and their higher rate of mental health issues prior to the bereavement, provides particularly robust evidence here. However, the picture is varied when other psychosocial outcomes are considered. One study found no significant difference between bereaved siblings and non-bereaved peers in relation to social measures and peer acceptance although there were sub-group effects (Gerhardt et al., 2012). This was a robust study (quality rating 83%), but relied on teacher report and peer rating to assess distress. It seems possible that many bereaved siblings may be able to continue to function in some spheres of life, but still have difficulties in others. Another study found that adolescent bereaved siblings' sense of self-concept was no different from normative levels (Martinson et al., 1987). This finding may be problematic given the high risk of bias in the study and especially the wide age range of bereaved siblings (8-18 years) in relation to a concept likely to differ according to age. However, this finding may reflect that self-concept is a stable construct and so may not change following bereavement.

In relation to the effect of time on bereavement, there is evidence that grief symptoms took more than a year to diminish and that grief continues to diminish for several years. This is consistent with findings relating to parentally bereaved children reported in a systematic review (Dowdney, 2000). In terms of long-term psychosocial outcomes, bereaved siblings appear to recover and measures of mental health indicated no significant differences from the wider population. Nonetheless, 2-12 years after death, few believed they had worked through their grief completely and on wider social measures sibling bereavement may be associated with some longer-term risks such as poorer schooling, an increased risk of dropping out of college and lower earnings in some circumstances (Fletcher et al., 2013). In summary, these findings are broadly consistent

with the trajectory of grief whereby people experience short-term problems followed by longer term resilience, but a minority experience longer term problems (Bonanno et al., 2002; Currier et al., 2007).

Factors Associated with Psychosocial Outcomes within Integrative Risk Factor Framework

Factors associated with poor psychosocial outcomes for siblings were explored within the context of Stroebe et al.'s (2006) framework. This confirmed that at least in the short term, death of a sibling is a loss-oriented stressor. Thus, two studies (Worden et al., 1999, Stikkelbroek et al., 2016) with quality ratings of 50% and 67% respectively, indicated that levels of distress of sibling bereaved children are equivalent to parentally bereaved children. In relation to the extent to which closeness of sibling relationship increases risk of poor psychosocial outcomes, the evidence is mixed and studies have a high risk of bias. Although any conclusion is tentative, it is possible that sibling bereavement is such a significant stressor that measurement of outcomes is not sensitive enough to detect gradations of closeness.

There was some evidence that restoration-oriented stressors (i.e. factors which are direct consequences of the circumstances around the death of sibling) affected psychosocial outcomes. Increased levels of involvement around the death and satisfaction with communication around the time of death were associated with better outcomes. This is consistent with qualitative literature which refers to the difficulties of not being involved in decision-making (Cross & Harrison, 2002; McNally, 2005). Although these results were consistent, the high risk of bias in relation to these studies mean that conclusions can only be tentative. No other restoration-oriented stressors were considered. Given known levels of parental distress and marital conflict after child bereavement (Rogers, Floyd, Seltzer, Greenberg, & Hong, 2008), this is a deficiency in the evidence-base relating to sibling bereavement.

In relation to intrapersonal factors, studies reported on the effects of age, gender and self-concept. In relation to age, the two most robust studies indicated that younger children may be more affected by death than adolescents. It would be helpful to have a better understanding of the mediating and moderating factors associated with this result (e.g. developmental understanding and levels of parental distress). Although one study

explored bereaved siblings' concept of death according to age, this did not consider the association with psychosocial outcome and so was not included within this review (Mahon, 1993). Exploration of other intrapersonal studies was limited. Bolton et al. (2016) explored whether sibling bereavement affected mental health problems when social economic status was taken into account, but no study specifically examined on the effect of social economic status on bereavement. In addition, no studies with validated outcome measures reported on factors such intelligence and religion.

Findings in relation to gender were also mixed with studies focused on the short-term measuring different outcomes, i.e. impact on social relationships (Gerhardt et al., 2012) and emotional and behavioural problems (Worden et al., 1999) meaning that results are difficult to synthesise. However, there was evidence that younger boys were more affected than girls regarding social relationships, but that girls had more internalising problems. In the longer term, Fletcher et al.'s (2013) study indicated that girls had longer-term effect in terms of education, teenage pregnancy and reliance on social support. These results seem to be consistent with evidence in relation to parentally-bereaved children that girls recover from grief more slowly than boys (Sandler et al., 2010).

Interpersonal factors include degree of social support or isolation and family dynamics. There was an association between anxiety and reports of social support immediately after the death of a sibling although conclusions drawn should be tentative because of the retrospective nature of these reports. The importance of family dynamics was also highlighted in the two studies which considered participants whose sibling had died in a homecare programme (Martinson et al., 1987; Mulhern et al., 1983). Despite the high risk of bias in these studies, these also provided evidence that communication and involvement in the sibling's death may be protective. In addition, consistent with findings in the literature relating to parental bereavement, parental distress was associated with sibling distress (Dowdney, 2000; Dowdney et al., 1999; Kalter et al., 2003).

In summary, the included studies investigated many of the risk factors within Stroebe et al.'s integrative risk factor framework. However, there was no evidence in the included studies in relation to coping mechanisms, cognitive processes and behaviour for sibling bereavement, except as regards the relationship between parental and sibling distress. In contrast, some qualitative studies explore these processes, for example Hogan and DeSantis (1992) and Packman et al. (2006) explored siblings' continuing emotional

and social bond with the deceased and Foster et al. (2009) explored the legacies of the deceased, identifying how such legacies could be motivational for siblings and their parents. This deficiency in the quantitative literature means that the current evidence cannot fully explain the processes by which risk factors contributed to greater problems.

Methodological Limitations of the Literature

Methodological issues also limit the literature reviewed. Although early studies in relation to sibling bereavement raised interest in the subject, they were also exploratory and failed to report hypotheses clearly. Furthermore, most studies had small sample sizes with participants from middle and upper social economic classes being overrepresented. This is especially problematic since studies indicate the majority of sibling deaths occur in the most financially disadvantaged families (Bolton et al., 2016). Despite recent studies of better quality (e.g. Bolton et al., 2016; Eilegard et al., 2013b; Morris et al., 2016), there are continuing issues. Most studies are cross-sectional or observational and rely upon retrospective reporting of events at time of death which increases risk of bias (e.g. Eilertsen et al., 2013; McCown & Pratt, 1985). In addition, several studies relied solely on parental report of sibling symptoms and there is evidence that parental perceptions of children's bereavement outcomes may not be a reliable measure (Applebaum & Burns, 1991; Hogan & Balk, 1990). Nine of the studies included more than one sibling in a family as an independent observer which is also problematic given evidence that psychosocial outcomes are similar across siblings in families (Birenbaum et al., 1989; Worden et al., 1999). Finally, the publication date of the studies ranged from 1983 to 2016 which means that there is a possible cohort effect with different generations having different psychosocial outcomes.

Strengths and Limitations

The strengths of this systematic review are that it was comprehensive, structured and protocol-driven with an explicit methodology. There were also limitations with this review in terms of selection criteria, screening and evaluation of the studies. Firstly, the decision to exclude studies primarily focused on bereavement occurring as a result of certain deaths (i.e. those before the birth of sibling, perinatal deaths, sudden infant death syndrome deaths, homicide and suicide) meant that the review did not explicitly explore the loss oriented stressor of how cause of death might influence psychosocial outcomes.

Secondly, related to the above limitation, over half the studies (Applebaum & Burns, 1991; Balk, 1983; Bolton et al. 2016; Demi & Gilbert, 1987; Fletcher et al., 2013; Hogan, 1988; Hogan & Greenfield, 1991; McCown & Pratt, 1985; Morris et al., 2016; Pettle Michael & Lansdown, 1986 and Stikkelbroek et al., 2016) either included such deaths or did not report the cause of death of the Index Child. This means that the psychosocial outcomes reported for bereaved siblings included the effects of these deaths, perhaps skewing the data. Thirdly, the selection criteria limited the review to studies which used a validated instrument to assess psychosocial outcomes; the review therefore omitted studies which describe sibling bereavement without formal instrumentation (e.g.Davies, 1991; Martinson & Campos, 1991; Rosen, 1986). Fourthly, only studies written in English were reviewed. Fifthly, as a result of limited time, the second reviewer only screened 20% of abstracts and titles, and full papers. Finally, although an attempt was made to evaluate studies for bias, the diversity of the methodologies meant that there was not a validated measure for evaluation and constraints in resources meant that there was no second reviewer.

Future Research

Examining the extant quantitative literature within Stroebe et al.'s integrative risk framework highlights many deficiencies in our knowledge about bereavement. Further research is necessary to investigate how family structures (such as marital breakdown and merged families) interact with sibling's experiences and impact psychosocial outcomes of bereavement. Given the association between lack of social support and communication, further research into the effects of participation in online social media support groups would also be helpful. Further quantitative research into cognitive and emotional processes which increase siblings' coping resources is also necessary to build on the current qualitative literature. However, it is possible that such research can take place within a wider context of investigating the effects of developmental stage on distress on children bereaved through death of a close family member.

Bereavement researchers need to ensure that where large cohort studies take place, data is captured which can inform understanding about sibling bereavement. Where studies cannot be longitudinal, they need to have control groups. This type of research will continue to be challenging. There is a potential reluctance of siblings and parents to participate in such research. There are also understandable concerns of clinicians and professionals about bereaved groups being approached. However, there is evidence from

the Swedish Study that contributing to research was a positive experience for participants (see Eilegard, Steineck, Nyberg, & Kreicbergs, 2013a)

Implications for Clinical Practice

It is important for clinicians and parents to be aware that sibling bereavement has a major impact on children's lives. In addition, although distress reduced over time, it will be helpful to recognise that it can take over two years for grief to lessen, but that eventually levels of stress seem to return to normal. In terms of naming those children most likely to be vulnerable to more complicated longer lasting grief and other problems, there is less evidence. However, clinicians and parents should be aware that younger children may be more affected than older children and that in the short-term boys seem to be particularly affected in their relationships with peers. There is also evidence that sibling death is more prevalent in the lower social economic class and that previous mental health problems are associated with greater psychosocial problems.

There is also some evidence that siblings who are less satisfied with the communication and social support during these times have worse outcomes both in the short and long term. Parents and clinicians should therefore be aware of the importance of communication and social support for bereaved siblings during the illness of the dying child, the death and the time after the death. This may include creating literature and online material that children can be directed to or offering consultations where children are able to ask questions.

Finally, parents and clinicians need to be aware of the associations between parental distress and sibling distress. Since Currier et al.'s (2007) review indicating that interventions for bereaved children are not effective, there is some evidence from a control group study that family bereavement interventions for parentally bereaved children are effective (Sandler et al., 2010). Such family approaches may also be effective in respect of bereaved siblings.

Conclusion

This systematic review summarises the extant quantitative studies of the psychosocial outcomes of sibling bereavement for children and young adults using validated instruments. It reveals many methodological problems with the literature, with most studies having a high risk of bias. Reviewing the literature in the context of Stroebe

et al.'s (2006) integrative risk factor framework highlights the need for more research in relation to the coping mechanisms, cognitive processes and behaviour for sibling bereavement. However, the literature suggests that sibling bereavement seems to follow the common trajectory of grief, with many siblings having worse psychosocial outcomes in the short term, but most returning to normal levels in the long term. Clinicians and parents should be made aware of the importance of communication with and social support for bereaved siblings and of the positive association between parental and sibling distress when faced with this very difficult event.

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Improving the Process for Referrals for Psychological Input: How Recovery Team Employees Make Decisions to Refer

Service Improvement Project

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Journal to be targeted

The Mental Health Review Journal includes work focusing on the delivery and evaluation of mental health services in the UK with particular attention to innovation, implementation and service user experience. Given this study's focus on the referral processes for psychological input, it falls within the remit of this journal (for details see Appendix 2.1)

Abstract

Psychological therapies are recommended by the National Institute of Health and Clinical Excellence (NICE) for many mental health problems, including anxiety and depression, yet resources are limited and not everyone is able to engage with therapy. Practitioners in Community Mental Health Trusts (CMHT) are often gatekeepers for access to psychological therapies, but little is known about how decisions to refer are made.

Martin (1999) suggested that clinical decision-making occurs in the context of three interacting elements a) the self; b) the organisation and wider structure; and c) the service user. To find out how CMHT practitioners refer to psychological therapies, practitioners within a CMHT were invited to take part in a study and 11 practitioners in a CMHT were interviewed about their decisions to refer to psychological therapies using a semi-structured interview schedule based on Martin's framework.

Results were analysed using thematic analysis resulting in eleven sub-themes under the three main themes of the self, the organisation and wider structure and the service user. Results indicated that some participants referred automatically if a service user asked or there was external pressure to refer while others' decisions were also informed by contextual information such as the service user's ability to engage or change, the risk presented and organisational limits in resources.

Recommendations were made for improvement to the service based on findings from these interviews.

Introduction

Psychological and psychosocial interventions are recommended by the National Institute of Health and Clinical Excellence (NICE) for a range of mental health problems including anxiety, depression, psychosis and personality disorders (NICE, 2005, 2009a, 2009b, 2011a, 2014). A systematic review also found that between 5% and 66% of patients with mental health problems preferred psychotherapy or counselling to antidepressant medicine (van Schaik et al., 2004). Yet not all service users are able or wish to engage in psychological interventions with studies indicating that they are less likely to complete therapy if they would have preferred medical treatment (Chilvers et al., 2001). Furthermore, resources for psychological therapies are limited; a King's Fund briefing found that most services do not have sufficient staff to provide NICE recommended interventions and that treatment waiting times for severe mental health problems are often over a year (Gilburt, 2015). This means that decisions need to be made about who should be referred to psychological therapies.

In England, people with serious mental health needs are usually supported in secondary care by community mental health trusts (CMHTs), multidisciplinary teams comprising social workers, mental health nurses, occupational therapists and doctors (Gilburt, 2015). Practitioners in these teams co-ordinate care and collaborate with service users to create care-plans covering their goals. Within this role, they have considerable flexibility and so may be involved in resolving housing and social care issues, referring to psychiatrists for medication reviews, as well as referrals for psychological therapy. Psychological therapies may be part of the CMHT, but service users do not usually have the option to self-refer. This means they do not have direct access to psychological therapies, but their care co-ordinators are the gatekeepers for psychological interventions.

Decision Theory

To understand how decisions are made about referrals for psychological input, it is helpful to understand the processes involved in decision-making. Information processing theory suggests that people are highly selective about what information we attend to and how it is used and we use heuristics (simplification mechanisms) to select and process information (Beresford & Sloper, 2008). Therefore, they only attend to factors that fall within a heuristic (e.g. relating to age, profession or race) rather than making decisions

based on all relevant factors. While heuristics are essential for quick decision-making, they can also lead to stigmatising biases whereby incorrect assumptions inform our actions. The dual process model of reasoning provides that individuals mostly use a combination of heuristics to select and process information, but on occasion they use complex cognitive processes (Eysenck & Keane, 2010). Kahneman and Frederick (2002) explain this in terms of two systems: the first system generates intuitive answers to judgement problems which are monitored by the second system which may correct them. In relation to clinical decisions, this means that people develop heuristics for making quick decisions and may not always consider relevant contextual information.

Decisions to Refer

Although dual processing theory gives an account of the processes involved in decision-making, it does not provide a framework for understanding the factors which influence decisions. Martin (1999) identified three inter-related concepts: the self, the service user and the social system, which influence the mental health nurses' clinical decision-making. In relation to self, nurses' judgements were found to be influenced by their attitudes, beliefs, values and changing emotional states. The service user's personality also influenced nurses' judgements while, in relation to the social system, the structure of the healthcare system and the physical and social environment impacted on decision-making.

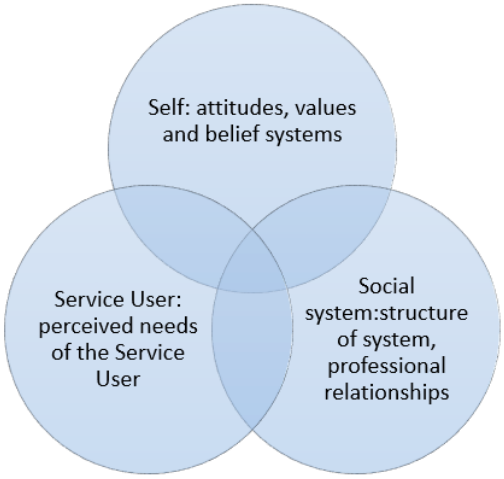

Figure 2.1. Model of clinical judgement. Reprinted from "Influences on clinical judgement in mental health nursing" by Martin (1999). *Nursing Times Research, 4*(4), 278. Copyright 1999 by Sage. Reprinted with permission.

Research which focuses on general practitioners' (GPs) decisions to refer to psychological services also highlights the interrelationship between factors relating to self, service user and social system. In relation to self, Fitch, Daw, Balmer and Gray (2008) have found that referrals are more likely if the GP is psychologically-minded. Referrals are also influenced by service user factors. Patients who are riskier and who evoked negative responses in GPs were referred more frequently (Knight, 2003; Sigel & Leiper, 2004). In a qualitative study exploring how 14 GPs made decisions to refer to psychological therapy, Stavrou, Cape and Barker (2009) found that three interrelated themes distinguished patients who were referred and those who were not. These were (1) patient initiative (i.e., where patients had specifically requested additional help in relation to their problems); (2) patients likely to benefit from psychological therapy; and (3) GPs' capacity to help. In addition, a background context impacted referrals to psychology. GPs were aware of psychology as a limited resource so they prioritised those they perceived would use the referral well and benefit most.

Few studies have specifically explored decision-making in mental health practitioners within CMHTs regarding the referral of service users for psychological therapy. However, Prytys, Garety, Jolley, Onwumere and Craig (2011) investigated CMHT practitioners' attitudes and decisions in relation to NICE recommended treatments for schizophrenia (NICE, 2014). They found an interaction between the factors relating to the practitioners themselves, the service user and the system. Practitioners were sometimes unsure of the value of psychological treatments for people with psychosis and were unable to articulate the trust referral criteria for psychological therapies or NICE guidelines, but they were guided by service users' views on treatment and limits in resources. This study highlights some of the complexities of the gatekeeper role.

In summary, mental health practitioners in a CMHT have an important and powerful role as gatekeepers for psychological therapies, yet little is known about how they decide to refer. This study aimed to understand how mental health practitioners decide to refer for psychological input using interview and qualitative methods and to make recommendations about how to improve the referral system in relation to that service based on the findings. Dual process theory and Martin's (1999) framework for clinical decision-making underpinned decisions about analysis and the recommendations made. The study sought to answer the following questions:

1) What factors relating to mental health practitioners' beliefs and attitudes, the service user and social systems, influence their decisions to refer for psychological input?
2) What do mental health practitioners find helpful and unhelpful about the referral process and methods for obtaining psychological input?
3) What further training can be provided about referrals and what changes to the process can be made?

Method

Description of the Service

The Complex Psychological Interventions service (CPI) in Bristol was commissioned in September 2014 and comprises professional qualified therapists, e.g. clinical and counselling psychologists, family therapists, psychodynamic psychotherapists and art psychotherapists (CCG, 2013/14). It has two main objectives, the first is to work directly with service users ensuring equal access to evidence-based treatment psychological interventions, a goal consistent with government strategy (Department of Health, 2011). The second is to support professionals within mental health services in implementing a Psychologically Informed Environments (PIE). The concept of PIE is rooted in a belief that service initiatives should be locally driven, arising from reflective practice within a service's staff team with a focus on the service user's psychological and emotional needs (Haigh, Harrison, Johnson, Paget, & Williams, 2012).

To meet the first objective, CPI offers a range of evidence-based therapies consistent with NICE guidelines e.g. cognitive behavioural therapy for anxiety disorders and depression (NICE, 2014), art therapy for those who have suffered psychosis (NICE, 2014) and dialectical behaviour therapy for people diagnosed with personality disorders (NICE, 2009a). To meet the second objective of supporting professionals in creating a PIE, CPI provides indirect psychological support and reflective space to professionals within mental health teams. Therefore, CPI professionals attend team meetings and local area meetings (stream meetings) where service users are discussed and provide regular supervision and formulation sessions.

Although CPI provides a range of evidence-based therapies, service users can only access these therapies if a referrer requests CPI input on an online form. This means that mental health workers are the gatekeepers to both indirect and direct psychological input since they decide which service users to discuss within meetings, formulation and supervision and which to refer for psychological therapies or for one to one advice. Five services can refer to CPI: the Recovery and Assessment Service (RAS), the Assertive Engagement Service, the Early Intervention in Psychosis Service, the Crisis Service and Inpatient Services. Most referrals derive from the RAS, which is equivalent to a CMHT and so the focus of this study is on decisions to refer to CPI by RAS professionals. Within the RAS there are two categories of mental health practitioners: Recovery Practitioners (RPs) who have a core health profession (e.g. social work, occupational therapy or mental health nursing) and Recovery Navigators (RNs) who are not usually qualified health professionals, but have experience of working in mental health.

Participants

RNs (n=14) and RPs (n=28) with a minimum of six months' experience within the RS were invited to participate in interviews via email, announcements and by one of the project supervisors (FN). Eleven participants were recruited, five females and six males. Six were RPs; of these, four were mental health nurses and two were occupational therapists. Five were RNs who had experience in mental health. Participants had a range of 9 months to 7 years' experience in the RAS.

Ethical Considerations

This study was reviewed and approved by the University of Bath Psychology Ethics Committee and the local NHS Trust Research and Development Department (R&D) (Appendices 2.2 and 2.3). All participants were given information before taking part in the study (Appendix 2.4). They were informed that they could withdraw their data at any point and request that data be destroyed. They were also informed that their employment would not be affected by participation in the study (or by a choice not to participate) and that their data would be anonymised. Interviews were recorded and kept in a locked drawer on a password protected memory stick. The first researcher transcribed all interviews, after which recordings were destroyed. Quotes were edited to facilitate reading and identifying details were changed.

Procedure

Interviews were conducted individually or in small groups. Six interviews were conducted: one with three participants, three with two participants and two were individual interviews. Interviews lasting between thirty minutes and an hour were conducted by the author in RAS meeting rooms. To ensure that the same topics were covered across interviews, a semi-structured interview schedule was used which was guided by Martin's (1999) framework for clinical decision-making (Table 2.1).

Table 2. 1

Questions for semi-structured interviews

	Questions and prompts
1.	How do you decide who to refer to CPI?
2.	In the cases of service users whom you do not refer, what helps you decide you are not going to?
3.	In relation to the referral structure and external systems – what makes it easy to make referrals? Are there things which make it more difficult? Could it be made easier?
4.	Is there anything about a service user that makes you more likely to refer? Do you ever discuss psychological therapies with service users to find out what they think? What kind of thing makes it less likely for you to refer?
5.	Who do you believe benefits most from psychological therapies? Are there some things or people you believe are just not appropriate for psychological therapies?

Position of the Author

The primary author, interviewer and data analyser (LF) was a trainee clinical psychologist who had spent six months on placement in a secondary mental health recovery team in a different part of the city. LF did not know any of the participants.

Analysis

The research was conducted from a realist perspective reflecting an assumption that the participants' spoken words in the interviews reflect their mental states and attributes, including meaning and intentions (Gorski, 2013; Potter & Wetherell, 1987). Interviews were analysed using a thematic approach as described by Braun and Clarke (2006), a

flexible approach to qualitative analysis which enables the researcher to focus on specific research questions. A deductive approach was taken so that the interviews were coded within a framework of themes imposed on the data, relating to (1) organisation structure and wider social systems, (2) service user qualities and (3) beliefs and attitudes to psychology based on Martin's (1999) study of community psychiatric nurses' decision-making. This top-down approach was selected because it enabled the researcher to ensure that data was organised to meet the aim of the project to provide recommendations to CPI about the decision-making process of mental health practitioners. It is consistent with the realist perspective and an analyst driven approach which gives a more detailed analysis of a specific aspect of the data described by Braun and Clarke. They state:

> *You can code for a quite specific research question (which maps onto the more theoretical approach)…*(Braun and Clarke, 2006 p. 84)

Interviews were transcribed verbatim by LF and read several times to increase familiarity with the data. Transcripts were then imported into NVivo-11, a professional software package designed to facilitate qualitative analysis. Coding was an iterative process with provisional codes identified and recorded in the coding scheme under the three overarching themes until saturation when all further text could be coded into the existing codes. Codes with similar meanings were then combined to form sub-themes with extracts being re-read to check that these were coherent and valid. Where themes were similar, they were merged and where they were inconsistent or lacking sufficient data they were discarded. The data-set was re-read to check that themes reflected the interviews. The NVivo software queries tool was used to analyse thematic patterns according to the role of the interviewee (i.e. RN or RP) or type of interview (group or individual). This enabled the researcher to consider whether some themes and experiences were more typical of an RP or RN role and whether participants responded differently to questions when interviewed collectively. To enhance analysis, Dr Kristina Bennert (KB), who has 20 years' experience in qualitative research, independently coded and generated sub-themes for 50% of the data. Themes were discussed and a final consensus was reached. Opinions about key meanings and themes were very similar between the researchers. As a final check, Dr Clare Sheahan, a lecturer in clinical community paediatric medicine, coded quotations from one interview. Inter-rater reliability was calculated using Cohen's kappa (k). The overall k value was 0.70 indicating substantial agreement.

Results

As described above, sub-themes were coded under three areas relating to decisions to refer to CPI: "Beliefs and attitudes about psychology", "Service user qualities" and "Organisational structure and wider social systems" (see Table 2.2).

Table 2. 2.

Number of participants coded for each theme

Main theme	*Sub-theme*	*N coded for theme*
Beliefs and attitudes	Most people could benefit	10/11 participants
about psychology	CPI is not always the answer	9/11 participants
	I want CPI when SUs are high risk	8/11 participants
	I'm not a psychologist.	7/11 participants
Service user (SU)	View of therapy	8/11 participants
Qualities	Stable enough to engage	9/11 participants
	Recognising need to change	9/11 participants
Organisational	External pressures	9/11 of participants
structure and wider	Indirect psychological input	11/11 participants
social systems	Uncertainty about acceptance criteria	4/11 of participants
	Limits in resources	11/11 participants

Beliefs and Attitudes about Psychology

Four sub-themes were identified relating to participant's beliefs and attitudes about psychology: (1) Most people could benefit; (2) CPI is not always the answer; (3) I want CPI when service users are high risk and (4) I'm not a psychologist.

Most people could benefit. Almost all participants expressed the view that most people could benefit from psychology even if therapy was not necessarily suitable at that time.

RN4: *In an ideal world, quite a lot of people* [would benefit from psychology] *really. That's why they come into the service because they need support with their mental health and their emotions.*

This extract illustrates many participants' view that psychology was key to the recovery service, although some distinguished between service users needing therapy from CPI and benefiting from it. Participants also identified when psychological input was not necessary.

CPI is not always the answer. In relation to this theme, some participants indicated that sometimes the focus of work was on the practical needs of service users:

RN1: *And it might be more about sorting out something a bit more practical with them, sorting out medication, housing, community activities, so on that sort of thing you wouldn't refer to CPI because there isn't a need*.

Other participants discussed how a service user may not be appropriate for therapy when they have already had therapy in the past. Another view was that, in their role as RPs or RNs, participants were providing a therapeutic relationship and therefore there was not always a further requirement for psychology. This is illustrated by RP1 below:

RP1: *Yeah, well we do it too. To a degree our work is that. Sometimes you've built a relationship with somebody and you think, "I'm doing the trick. You know, there's no point, really*"

Here the RP asserts his own value as a support for service users and two other participants, one RP and one RN of considerable experience also reflected this view.

I want CPI when service users are high risk. Participants speaking about seeking CPI support when service users were at high risk were aware that such people were not necessarily the people who would be able to engage in therapy.

RN4: *They might not fit the criteria for psychology, but it would be really helpful to have psychology discussion because you are really struggling.*

This extract indicates that indirect psychological input was valued as a support for participants. Yet two participants expressed their frustration that those at most risk were seldom accepted for therapy.

RP6: Ummm… although the rational part of me is like, yeah, "that work probably wouldn't be helpful right now." When you have somebody who is so complex and has all these needs, then it's frustrating.

I'm not a psychologist. A further sub-theme was the belief that CPI had specialist knowledge and so there were certain decisions which it was not appropriate to make.

RN2: *I don't know what you should have and that's why you would have an assessment with a psychologist and sort of talk about that there, you know. Because I wouldn't pretend to know which therapy you should have or anything.*

Although some participants, notably the OTs and the RPs who had been within the service for a longer time, had some knowledge about the therapies available, they also expressed the view that the ultimate responsibility lay with psychology.

RP4: *Because ultimately, it's got to be CPI's understanding of what is useful for the person*.

Service User Qualities

In relation to service user's qualities, three sub-themes were identified: (1) View of therapy; (2) Stable enough to engage; and (3) Need for change.

View of therapy. The service user's view of therapy was a significant factor in whether participants referred them to therapy.

RN2: *I think from my point of view, if someone asks me then I do. I have not come across someone where they haven't asked and I've thought, "They really need it yet." … They probably say, "I could probably do with some therapies or talking therapies" or whatever else they ask for, I will then talk to CPI.*

For this participant, a service user's request for therapy was necessary for them to make a request for input. Other participants took a more active role, sometimes suggesting to a service user that they might want to consider therapy or deciding not to refer even when the service user asked:

RP6*: Yeah, so if [the service users] are asking, asking, asking. And I don't think this is appropriate and that's what I'm feeding back. But then ultimately, I just actually want to talk to* [CPI].

This extract indicates that participants do not always accept a service user's view of therapy as an overriding factor, but this can be an uncomfortable position for practitioners who then prefer to discuss with CPI.

Stable enough to engage. Almost all participants discussed the importance of service users' ability to engage, highlighting that other aspects of service users' lives needed to be relatively settled.

RP4: *I have had conversations with CPI as well where they've talked about it, you know kind of other factors like being in stable accommodation and that sort of thing to start long term therapy if someone is homeless or in short term accommodation it is not usually a good time to start then.*

Many participants indicated that if a service user could not engage with them, they were unlikely to refer to therapy:

RP2: *Well, there are people that we find it hard to engage with and that's us going out to houses, GP surgeries, community centres… it's that those types of service users, I really wouldn't refer to psychology.*

Recognising need for change. Many participants spoke about referring only when participants were aware that they needed to change. This was sometimes framed as psychological insight.

RP2: *They need insight to begin with… Yeah. An awareness that shifting something in them can make a change, rather than wanting the world to change, or other people to change.*

Participants also discussed how service users' readiness to change affected both their decisions to refer and the course of therapy.

RN2: *I think it's the individuals who want the most change who get the most out of it.*

In contrast, participants spoke of service users who were "stuck". The extract below illustrates the view that service users who were not ready to change were not suitable for therapy.

> RN4*: No. And it's hard because…you are permanently thinking that maybe you will someday be ready for this. But they are really stuck and going round in circles and have been for a long time.*

Organisation Structure and Wider Social Systems

In relation to how structure of the service and the wider social system influenced referrals to CPI, four distinct sub-themes were identified: (1) External pressures to refer; (2) Indirect psychological input; (3) Uncertainty about the criteria for acceptance for psychological therapies; and (4) Limits in resources.

External pressures. Participants reported that referrals for therapy were driven by professionals and family members outside the recovery team. They indicated that when this occurred they often automatically referred and so although they completed the paperwork to refer, they did not see their role as decision-makers in the process. The extract below exemplifies this approach.

> *RP2: Sometimes people have come with a referral to psychology. And they might have already had some sort of psychology, like IAPT and they've said that they need more intensive, more ongoing therapy or be suicidal or whatever.*

All participants indicated that they referred to CPI in these circumstances, yet some expressed frustration at the pressure from psychiatrists or family members, noting that the service user may not be appropriate for therapy.

> RN4: *But sometimes there is pressure as well from sort of doctors' reviews. So, there have been some [psychiatrists] who say, "Yes, psychology, psychology...." No one here at the moment, but there has been almost promises to the SU.*
> LF: *And is that a difficult thing?*
> RN4: *Yeah, because it's kind of a process. It's not like we refer to psychology and they say yes. It takes time and there's criteria.*

The extract above illustrates how the actions of other professionals could sometimes raise service users' expectations that they would receive therapy which participants could not always meet because CPI would determine service users' suitability for therapy.

Indirect psychological input. The system for obtaining indirect psychological input had changed in the year before the interviews when the PIE model was introduced. Most participants indicated that they sought input from CPI without making a formal request by using the structures and systems of the recovery team. A typical example is demonstrated in the extract below:

> RP2: *I would bring it up in stream* [meetings]. *Because if you are referring somebody for psychology then you have got concerns about their wellbeing anyway and what's best for them.*

Although many found these systems helpful and preferred using them to arranging one-to-one meetings, other participants expressed reservations:

> RP6: …*we also have another way, I suppose, of having conversations which is in our stream meetings. Which I don't feel very helpful to be honest.*
>
> LF: *Do you know why not?*
>
> RP6: *There's too many people in the room. Not enough time to really think about it…*

This is an indication that a uniform approach to obtaining indirect psychological input was unlikely to be appropriate. Working hours, preferences for type of advice and personality all influence decisions to refer to CPI.

Uncertainty about acceptance criteria. Many participants conveyed their uncertainty about criteria to be accepted into psychology.

> RN3: *I think myself and a lot of recovery navigators are still a bit hazy about the criteria for acceptance and whether someone is going to get on to CPI or not… there's historically been thresholds for not complex enough and too complex, or too unstable and trying to get within that band of acceptance has been quite trying (laughs).*

This was also a problem faced by at least two RPs:

> *RP6: I guess nobody has ever said... I've made up in my mind, it's entirely me whose made up who's going to benefit from psychology.*

The issue of lack of specific information about CPI acceptance criteria was exacerbated by participants' concerns regarding limits in resources.

Limits in resources. Almost half the participants discussed how limits in resources affected their decision-making:

> RP5: *For me yeah, because I just think, I want to make sure I'm getting the most prioritised people into it and want to make sure if I am getting somebody into it, it has to be somebody who really needs it.*

In this extract, RP5 acknowledged that limits in resources might affect his decision-making, but indicated that often it would not. Other participants reported that their decision-making was also affected by gaps in the structure of services, which was a key concern.

> *RP4: There are still people who perhaps might slip through the net almost because, they are not risky enough or not that much in need of therapy as some other people on our case load, but they are too unwell to see wellbeing therapies. And so, there's a gap.*

Discussion

The study aimed to understand how RNs and RPs in an RAS made decisions to refer and access psychological input within Martin's (1999) framework.

Beliefs and Attitudes to Psychology

Almost all participants saw psychology as potentially valuable for service users, but their beliefs and attitudes about psychology in the context of limited resources drove decisions about referral. Participants reported prioritising risky clients for psychological input because they needed emotional support. This means participants consciously chose to talk about service users who were high risk. This is similar to the approach of GPs who

referred to psychology where they thought that supporting the service users themselves was beyond their capabilities (Stavrou et al., 2009). However, for low risk service users, participants commented that there was not always a need for psychology because they were capable of "doing the trick". This may be an unforeseen consequence of the PIE, which emphasises all staff reflecting on the psychological and emotional needs of the service users, but does not necessarily privilege evidence-based therapies. This is a potential concern because people who do not present as higher risk are less likely to be referred even when potentially suitable for evidence-based therapies.

A sub-theme here was the view that RNs and RPs were "not psychologists" and therefore ought not to decide which treatments were beneficial. Although some participants had some idea of what psychological therapies were appropriate for particular service users, no participants referred to NICE guidelines about access to psychological therapy. This reflects that participants did not consider the evidence base in relation to decisions to refer. While RNs and RPs would not ultimately make decisions about which therapy would be right for a service user, it seemed important that they had an overview of evidence-based therapies to be effective. This issue was highlighted as an area for potential improvement for CPI.

Service User Qualities

Service user qualities were also important in participants' decisions to refer. Almost all participants stressed that a service user requesting therapy was important to decision-making. Indeed, for some people, the service user asking for therapy or external pressures to refer were the only drivers to refer. The service user's agency as a decision-maker in their treatment is emphasised within NICE guidance (NICE, 2011b) and has been shown to be an important factor in treatment outcomes (Chilvers et al., 2001). However, if recovery team members rely solely on service users requesting treatment, some will not be provided access to NICE recommended treatments and others unsuitable for referral will be referred.

For other participants, factors about the service user, beliefs about the service and their own beliefs interacted, meaning their decisions to refer were more contextually based as described in the second route outlined in dual process theory (Eysenck & Keane, 2010).

In these cases, participants took into account not only the limited resources, but also service user's ability to engage in therapy and openness to change.

Organisational Structure and Wider Systems

Decisions to refer for psychological input were made in the context of the wider organisation and structure. Participants spoke of using team meetings, team formulation sessions and supervision to seek psychological input instead of making formal referrals for psychological input. This indicated that, in accordance with the concept of PIE, psychology was embedded within the RAS (Haigh, Harrison, Johnson, Paget, & Williams, 2012). However, it also suggests that the creation of a PIE may sometimes result in fewer formal referrals for psychological input and therefore service users not receiving evidence-based psychological therapies, but receiving psychologically informed care as a result of being discussed with professionals within CPI. However, it was also apparent that some decisions about whom to refer were not made as a result of the PIE, but on the basis of psychology being a limited resource and it being necessary to prioritise some service users over others. This is consistent with the King's Fund briefing finding that few services had capacity to provide recommended NICE treatments (Gilburt, 2015).

The issues raised in the two sub-themes, (1) External pressure to refer and (2) Uncertainty about criteria for referral, highlighted areas for potential improvement for CPI. Some participants would automatically refer to psychology if there were external pressures to refer. In the context of the dual process model of decision-making, this meant that they had an intuitive response (e.g. "I refer when requested by the psychiatrist") without taking into account wider contextual information (Kahneman & Frederick, 2002). In these cases, practitioners were no longer acting in a gatekeeper role. It was important that CPI became aware of this practice so that professionals could advise RNS whether this was the correct approach.

Several participants expressed uncertainty about the criteria for referral. This is consistent with Prytys et al.'s (2011) findings that practitioners were often unable to articulate trust referral criteria. Participants also stated that previous refusals of referrals for therapy could make them reluctant and uncomfortable to refer to CPI again. This was an important issue to highlight to CPI since this uncertainty could mean that some people appropriate for therapeutic support were not referred.

Summary of decision-making for referrals to psychology

In summary, sub-themes under the main themes of organisation structure and wider social systems; service user qualities; and beliefs and attitudes to psychology had varying impact upon referrals to psychology and these can be understood in the context of information processing theory. Service user view of therapy and external pressures to refer were key to the referral process with some participants indicating that without these factors they would not make a referral, but in their presence, they would always refer. This is indicative of use of the first system within the dual processing model for quick intuitive decisions using a heuristic (Kahneman & Frederick, 2002). However, other participants employed the second system, taking into account contextual information before making a decision. Five themes related to the contextual information influencing decision making: the service user being "stable enough to engage" or recognising "a need to change"; the participant recognising that psychological "resources are limited" and therefore wanting to have psychogical insight on "the most risky service users"; and a belief that "CPI is not always the answer" especially if participants believed that they were "doing the trick". In contrast, although the themes that "most people could benefit from psychological therapies"; that it was possible to obtain "indirect psychological input" and that there was "uncertainty about acceptance criteria" for psychological therapies were relevant to the decision-making, they did not appear to either increase or decrease referrals to psychology. Figure 2.2 below sets outs a simplified model of how decision-making may be made.

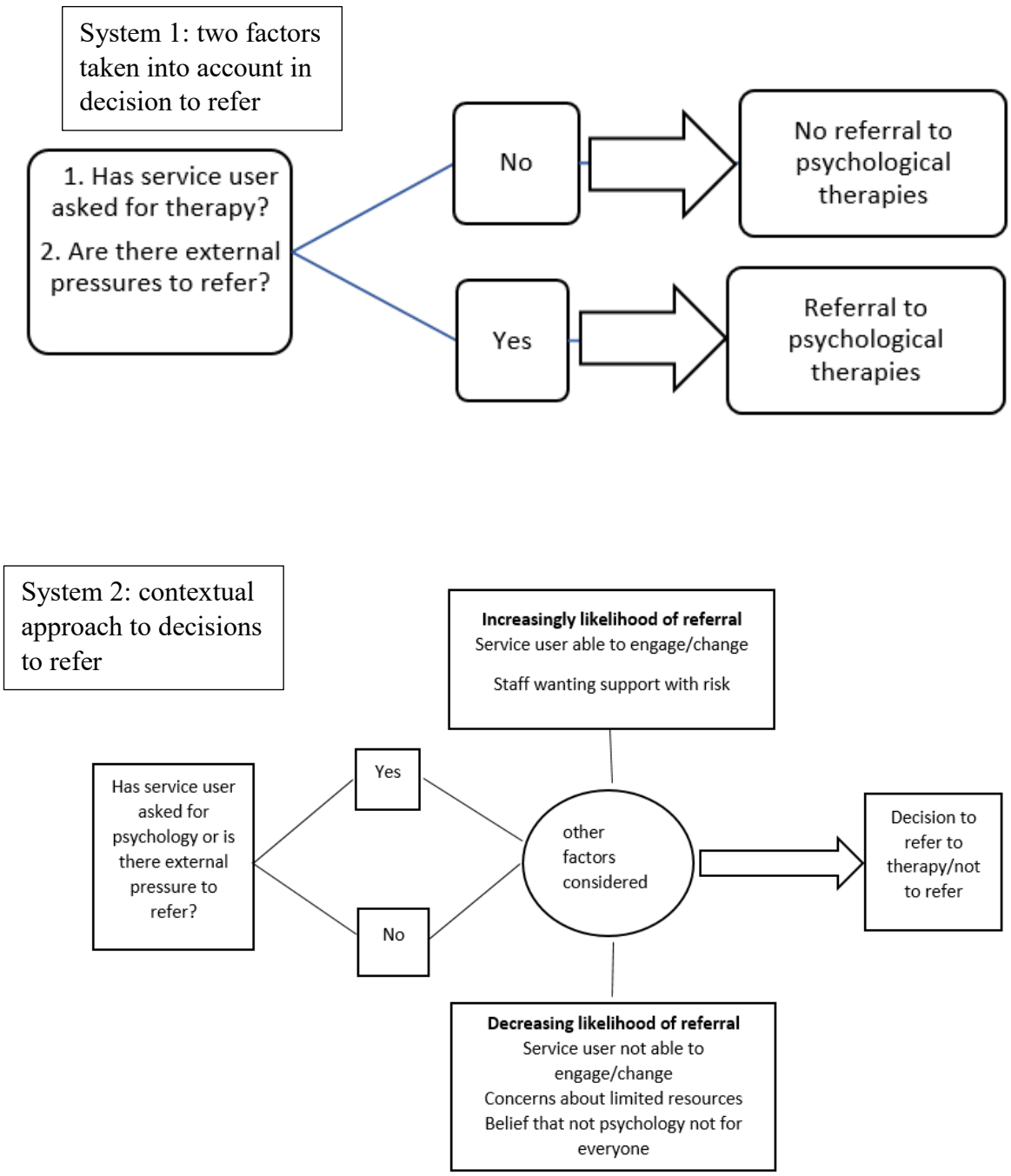

Figure 2. 2. Model of decision-making depicting system one and system two processes

Recommendations

LF attended a CPI local meeting to present findings and discuss how recommendations could be implemented in the service context (see Appendix 2.5). At the meeting, the consensus view was that it was not the role of RNs and RPs to make decisions about type of psychological treatment; but that it was important that RNs and RPs took contextual issues into account to ensure those who would benefit from psychological input could gain access rather than those who held the most risk. The meeting also revealed a lack of clarity amongst CPI about criteria for referral. A second service improvement in a different area of the service had also highlighted that, although there was a constant demand for training, this was often ill-attended because of shift patterns and workloads. Furthermore, a three month follow-up of one training session on different types of therapy had revealed that RPs and RNs gained understanding in the session, but did not retain this information. Recommendations to the service took this feedback into account in considering how information could be conveyed through means other than formal training. Following this meeting the recommendations on how the service might be improved were circulated. These are summarised in Table 2.3.

Limitations

There are several limitations to this study. The original intention was to conduct focus groups for recovery practitioners and recovery navigators. However, it proved impossible to arrange meetings for larger groups because of conflicting work schedules. Therefore, interviews were conducted both with individuals and with groups of one and two and it is possible that people may have shared different views when talking within a group rather than as an individual.

In addition, participation was voluntary. Participants were encouraged to participate by FN who was a clinical psychologist and member of CPI and participants were aware that LF was training to be a psychologist. This could have resulted in a sample who expressed a more positive view of the role of psychology than others who did not choose to be interviewed. However, within the group interviewed, there were both occupational therapists and psychiatric nurses and there was also considerable variation in experience of working in mental health and within the team. The interviewer did not know any of the participants and they did seem to be comfortable talking openly.

Table 2.3.

Summary of recommendations shared with the team

Area	*Suggestions*	*Next steps*
External pressures to refer	Clarity provided to RNs and RPs as to gatekeeping role in relation to external referrals.	CPI to consider at a meeting whether external pressures should lead to automatic referrals.
SU requests	Clarity provided to RNs and RPs as to gatekeeping role in relation to SU rather than simple heuristic (they ask, I refer, they don't ask I don't) (see also NICE guidelines below).	CPI to create working group to create guidelines about referrals to CPI?
NICE guidelines	NICE guidelines were not mentioned by any participants. Therefore, we recommend that NICE guidelines are discussed and so RPs and RNs are aware of NICE recommended treatments for those with less risk and that for certain problems, psychological treatments should be offered (e.g. for psychosis) (NICE, 2014)	Creating a table of NICE recommended treatments for different problems which is presented at team meetings and local area meetings and easily available on the service's intranet.
Uncertainty about criteria for acceptance by CPI	Establishing a key list of issues to be considered with every SU in relation to psychological therapy. This will need to be agreed by CPI as a group. But should include: stability, engagement issues and characteristics of disorder.	List created to be agreed at CPI meeting. This list to be attached to the referral for input form. If someone makes an inappropriate referral. They should be given this information.

Recommendations for Future Research

This research has highlighted that RNs and RPs often make referrals at the instigation of other professionals (e.g. psychiatrists, general practitioners and the primary care liaison team). Future research to understand the decision-making processes of such professionals would therefore be valuable. It would also be useful to monitor the impact of any changes in referral practice. This will reveal specific factors that may be effective at improving the referral process to CPI.

Conclusion

In conclusion, many different factors influence decisions to refer for psychological input in relation to RPs and RNs themselves, services users and the context of the service.

For some participants, the only influencing factors were service user request and external pressures to refer, while others considered some contextual factors including their own needs for support with risk, service users' ability to engage and change and the context of the service and limited resources. Recommendations for service improvement include providing clarity on the gatekeeping role of RPs and RNs and criteria for acceptance into psychological therapies as well as greater emphasis on NICE recommendations for treatment. Future research could focus on the decision-making of professionals employed in other services that refer to CPI.

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Guilt and Shame in Perinatal Death: a Comparison of Childless Women and Women who have Children

Main Research Project

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The Journal is a source of published material on the psychological aspects of human reproduction. It reports and reviews research on psychological, behavioural, medical and social aspects of human reproduction, pregnancy and infancy.

(for details see Appendix 3.1)

Abstract

In 2014, approximately 1% of births in England and Wales resulted in perinatal death, defined as stillbirth at more than 24 weeks of gestation or neonatal death of a baby within 28 days of birth. Of women who experience perinatal death, 15-25% have clinically significant mental health symptoms a year later which can be conceptualised as complicated grief. Shame and guilt-proneness are associated with prolonged grief in women who have experienced perinatal death, yet it is not known whether shame and guilt are triggered by perinatal death, or the risk factors for increased shame and guilt. This cross-sectional study explored whether state shame and guilt are associated with long-term grief even when controlled for depression. It also considered whether childlessness before perinatal death predicts feelings of state shame, and having living children predicts state guilt.

Three groups of women recruited online through social media and support groups completed a survey online or by telephone containing measures of shame and guilt-proneness, state shame and guilt, and grief. Groups comprised: women who had experienced perinatal death 13-36 months previously divided between participants with surviving children pre-bereavement (n=40) and participants with no children pre-bereavement (n=55); and a control group who had given birth to a surviving baby 13-36 months previously (n=96).

Bereaved groups reported significantly higher levels of state shame and guilt than the control group despite comparable levels of shame and guilt-proneness across groups. There were no significant differences between state shame, state guilt or grief within bereaved groups. A Pearson product-moment correlation indicated that state shame explained a significant proportion of variance in grief when controlled for depression (17%).

Understanding the role of shame when working clinically with women who have experienced perinatal death and developing interventions to reduce shame and guilt is an important area of research.

Introduction

In 2014, approximately 1% of births in England and Wales resulted in perinatal death, defined as stillbirth at more than 24 weeks of gestation or neonatal death of a baby within 28 days of birth (Office for National Statistics, 2016). Yet perinatal death is often viewed differently from other bereavements with qualitative studies indicating that parents sometimes believe that their grief is minimised by friends, relatives and healthcare professionals who view a perinatal death as a less traumatic death than that of a partner or older child (Brierley-Jones, Crawley, Lomax, & Ayers, 2015; Cacciatore, 2010; Lang et al., 2011). This is supported by a recent systematic review which found that disenfranchised grief was the second most common theme (after negative psychological symptoms) across studies (Burden et al., 2016). As a result, parents may not be supported in their grief.

Impact of Perinatal Grief

Perinatal death can cause prolonged grief comparable to death of an older child or partner (Burden et al., 2016; Giles, 1970; Nicol, Tompkins, Campbell, & Syme, 1986). Most parents have grief reactions with cognitive, emotional, physical and interpersonal disruptions including denial, sadness, shock and sleep problems (Bonanno et al., 2002; Boyle, Vance, Najman, & Thearle, 1996). Badenhorst, Riches, Turton, and Hughes (2006) reports in a systematic review that the pattern of grief for mothers and fathers who experience perinatal death tends to be similar, but women's symptoms of distress measured by anxiety, low mood and depression have been consistently reported as longer lasting and more intense. Most women (between 51 and 87%) adapt to the death without significant mental health disorders. However, in the first year they have increased risks of anxiety, depression and post-traumatic stress disorder and two years after the death between 15% and 25% have ongoing problems (Boyle et al., 1996; Engelhard, van den Hout, & Arntz, 2001; Radestad, Steineck, Nordin, & Sjogren, 1996; Turton, Hughes, Evans, & Fainman, 2001). Therefore, the psychological impact of perinatal death is an area in which better understanding is required.

Factors Associated with Poorer Outcomes

Several studies have investigated factors associated with poorer outcomes for women bereaved through perinatal death (for reviews see Badenhorst & Hughes, 2007; Bennett, Litz, Lee, & Maguen, 2005; Campbell-Jackson & Horsch, 2014). Higher levels of distress are associated with poor levels of social support, more recent bereavement and pre-existing mental health problems (Cacciatore, Rådestad, & Froen, 2008; Cacciatore, Schnebly, & Froen, 2009; Forrest, Standish, & Baum, 1982; Janssen, Cuisinier, de Graauw, & Hoogduin, 1997; Toedter, Lasker, & Alhadeff, 1988). Since 45% of women who experience perinatal death already have a child and between 50% and 80% have a subsequent child (Blackmore et al, 2011; Gordon, Raynes-Greenow, McGeechan, Morris, & Jeffery, 2013), understanding women's distress in relation to the perinatal death in the context of their family situation is important.

There is evidence from a longitudinal cohort study that histories of prenatal miscarriages and stillbirth predict greater anxiety and depression in pregnancy and after the birth of a subsequent child than women who have not had such experiences (Blackmore et al. (2011). However, when comparing women who have had stillbirth, the evidence in relation to the effect of pregnancy on grief and distress is mixed. In an online survey of 2,292 women who had experienced death of a baby of 20 weeks' gestation, Cacciatore et al. (2008) found that being pregnant after a death was associated with fewer depressive symptoms, but that there was not a significant effect in relation to anxiety.

There are also mixed findings about the impact of having living children before experiencing perinatal death. In a prospective longitudinal study using interview methods, Janssen et al. (1997) assessed 227 women who had experienced perinatal death and found that having living children before the death was associated with lower levels of grief. Similarly, in the online survey referred to above of 2,292 women who had experienced death of a baby of 20 weeks' gestation, Cacciatore et al. (2008) found that having living children before experiencing perinatal death was associated with lower levels of anxiety and depression. In their study of 194 participants, Toedter et al. (1988) found that this effect did not quite reach significance ($p<.55$) and suggested that any effect may be explained by age because older women had lower grief scores. Conversely, in a postal study of 314 women three years after stillbirth Surkan, Rådestad, Cnattingius, Steineck, and Dickman (2008) found rates of depression increased twofold where stillbirth occurred

in a third pregnancy compared to a first pregnancy. This means that no firm conclusions can be drawn about whether having a child before experiencing perinatal death is a protective factor for long-term distress. It is therefore also helpful to consider the psychological processes involved in grief associated with perinatal death.

Guilt and Shame Processes

Studies have highlighted that guilt and shame play an important role in grief with women being particularly vulnerable to more feelings of guilt (for review of the role of guilt see Li, Stroebe, Chan & Chow, 2014). Although the terms are often conflated in research into grief where the focus is often on guilt (e.g. Li, Stroebe, Chan, & Chow, 2014; Stroebe et al., 2014), Lewis' pioneering work distinguished the two emotions (Lewis 1971 as cited in Tangney & Dearing, 2002). Guilt and shame are both negative, self-conscious, moral emotions which occur as a result of transgressions or failures (Tangney & Fischer, 1995). Shame is described as an emotion focused on the self, while guilt is focused on behaviour (Tangney & Dearing, 2002). An individual who feels shame considers her global self to be at fault, while an individual who feels guilty may find fault with her behaviour, but does not denigrate her whole being. The phenomenological response and associated actions are also different: an individual who feels shame is likely to feel powerless, hopeless and dejected leading her to hide, escape or become angry. In contrast, an individual who feels guilty will often feel agitated, regretful and remorseful, which may lead her to make reparations. Tangney and Dearing (2002) also distinguish state shame and guilt, which refer to feelings of shame or guilt experienced in specific situations and shame-proneness or guilt-proneness which are innate dispositions to experience shame or guilt.

There is evidence that shame-proneness is associated with several psychopathologies including depression, substance abuse and post-traumatic stress disorder (Beck et al., 2011; Harper & Arias, 2004; Kim, Thibodeau, & Jorgensen, 2011; Meehan, O'Connor, Berry, Weiss, & Acampora, 1996; Tangney, Stuewig, & Hafez, 2011). In relation to women bereaved through perinatal death, there is evidence from qualitative studies that self-blame and guilt are key themes for some women who have experienced perinatal death (Cacciatore, DeFrain, & Jones, 2008; Goopy, St John, & Cooke, 2006; McCreight, 2008) and that shame is a key psychological process in disenfranchised grief of women who experience stillbirth (Golan & Leichtentritt, 2016).

In recent years, the roles of guilt and shame in prolonged grief reactions in perinatal death have been investigated empirically in quantitative studies (Barr, 2004, 2012; Barr & Cacciatore, 2007; Cacciatore, 2013; Cacciatore, Froen, & Killian, 2013). Barr and Cacciatore (2007) studied the relationship between personality proneness to problematic social emotions of envy, jealousy, shame and guilt and maternal grief in 441 women bereaved through miscarriage, perinatal death or child death. They found that chronic guilt controlled for envy and jealousy contributed to variance in grief but shame did not. In a longitudinal study, Barr (2004) studied the association between guilt and shame-proneness and perinatal grief for men and women at one month (Early Grief) and thirteen months (Late Grief) after the death of a baby. He found that shame-proneness, but not guilt-proneness explained a small variance in Early Grief in women (9%). However, 13 months later, shame-proneness explained a substantial proportion of the variance (27%) in women's late grief. Guilt-proneness controlled for shame made a further significant contribution (21%). In total, shame and guilt accounted for 45% of the variance in Late Grief in women.

Although Barr's study highlights that shame and guilt are important within perinatal death, it leaves several issues unresolved. Since it measures shame and guilt-proneness and chronic shame and guilt, it does not specifically investigate whether current feelings of shame and guilt are triggered by the perinatal death. In addition, it does not explore whether, if depression is controlled for, the association with perinatal grief with shame remains. This means that further investigation into the reasons women who have experienced perinatal death may experience such feelings and what predicts these feelings is important.

There is anecdotal evidence from clinicians working with women bereaved through perinatal death that women who have not had living children prior to experiencing perinatal death appear to have a greater sense of shame and women who have had children prior to the experience have a greater sense of guilt. There is only very limited theoretical justification for this observation, but it is possible that this sense of shame may be related to sense of denial of maternal identity. In an experimental study, Ferguson, Eyre, and Ashbaker (2000) found that feelings of shame are related to sense of identity. Brierley Jones et al. (2015) found that some mothers bereaved through perinatal death reported denial of their identity as a mother as part of their grief experience. In addition, a study by

Wonch Hill, Cacciatore, Shreffler and Pritchard (2016) indicated that the impact of perinatal death on self-esteem was moderated by maternal identity. Although there is no evidence specifically on this issue, a comparison of infertile couples with couples who had a healthy child, indicated that infertile couples had significantly more feelings of shame (Galhardo, Pinto-Gouveia, Cunha, & Matos, 2011). Furthermore, Schwerdtfeger and Shreffler (2009) found that of women who had suffered pregnancy loss and had not had children, 45% felt that they were inadequate and 36% that they were failures as women compared to women who had suffered loss and had children of whom 24% felt inadequate and 19% felt like a failure as women, indicating high levels of global shame (no effect sizes reported).

There is also little theoretical evidence in relation to why women who have children might feel more of a sense of guilt. However, in a qualitative study, women interviewed referred to their guilt and regrets in relation to their surviving children including failing to include them in rituals in relation to the baby, but also a concern that the death lead to questioning their ability to be a good mother (Cacciatore, 2010). Avelin (2011) also suggests that where mothers have surviving children, they can feel guilty in relation to their family because of not being able to do things that they should do for their children because of their grief. This could also provide an explanation why some studies have shown that people without living children have higher levels of distress, since it is likely that shame experienced by people without living children is more associated with grief and depression than guilt.

Rationale and Hypotheses

The present study is a speculative study prompted by the anecdotal reports from clinicians who have worked with women bereaved through perinatal death. They have commented that guilt and shame appear to be important emotions in grief, but also that women who have had children prior to experiencing perinatal death seem to have more guilt and women who have not, more shame. There is currently little theoretical evidence to support this observation, although there is some qualitative evidence which indicates guilt and shame may be experienced differently by women who have living children at the time of the perinatal death compared to those who have not had children. Although there is evidence that guilt and shame-proneness are associated with grief reactions in Late Grief

(Barr, 2004), there is no evidence in relation to whether state shame and guilt are triggered by perinatal death and memories and reminders of the baby. Investigating the relationships between shame, guilt and distress and whether having living children or not prior to perinatal death affects the experience of self-conscious emotions could enable clinicians to work with such feelings in a targeted way in the future.

The primary aim of this study was to investigate whether feelings of state shame and state guilt in Late Grief (between 13-36 months after perinatal death) are predicted by whether a woman has had living children prior to experiencing perinatal death. The secondary aim was to extend Barr's (2004) study and consider whether state shame and guilt triggered by recalling experiences of perinatal death is associated with grief controlled for depression. Hypotheses are outlined in Table 3.1 below.

Table 3. 1

Hypotheses

	Hypotheses
1	Women who have suffered perinatal death will report more state shame and guilt than women who have not suffered perinatal death.
2	Women who have suffered perinatal death and have at least one living child will score more highly for state guilt and less highly for state shame than women who have suffered perinatal death and have no children.
3	There will be no difference between women who have suffered perinatal death and women who have not suffered perinatal death in terms of guilt or shame-proneness.
4	Women who have suffered perinatal death will have higher scores for anxiety and depression than those who have not suffered perinatal death.
5	Women who have at least one living children before they experience perinatal death will have lower scores for anxiety and depression and grief than those who have no children.
6	State shame will be associated with perinatal grief in women who have experienced a perinatal death when the effects for depression are controlled.

Method

Ethics

The study was granted full ethical approval by the University of Bath Psychology Ethics Committee (16-229) (Appendix 3.2). Women with personal experience of stillbirth were consulted in the design stage and piloting of the study and amendments were made to reflect their comments. SANDS-UK, the stillbirth and neonatal death charity also reviewed the study and before advertising on the SANDS page online, they requested that the survey be as brief as possible, the word death instead of loss be used and that no specific prompts were used to ask participants to think of their baby.

Participants

Three groups of adult female participants were recruited (N=191). There were two bereaved groups comprising women who had experienced perinatal death of a baby between 13 months and three years previously: the bereaved sample (Bereaved-F) had not given birth to a living child before the death of their baby (n = 55); the bereaved group with children (Bereaved-C) had a living child born before the perinatal death (n = 40). A third group, the control sample (Control), comprised women who had given birth to a baby between 13 months and three years previously and had never experienced perinatal death of a baby (n = 96).

Inclusion criteria were: 1) being female, 2) aged 18 years or older, 3) ability to read English. Exclusion criteria for the bereaved samples were: 1) perinatal death being a consequence of late termination of pregnancy for medical reasons; 2) multiple birth in which one child survived and the other(s) did not; and 3) perinatal death 13 months before completing the survey.

Design

A mixed design was employed for this study. A between-groups causal comparative questionnaire design investigated differences in questionnaire responses. The independent variable was a participant group comprising three levels (Bereaved-F group, the Bereaved-C group and Control group) and the dependent variables were shame-proneness, guilt-proneness, chronic shame, chronic guilt, state shame, state guilt, depression and anxiety.

A within-groups comparative questionnaire design investigated associations between measures of shame and guilt, grief, depression and anxiety in relation to the bereaved participants only.

The state shame and guilt questionnaire was intended to assess shame and guilt triggered by reminders of perinatal death. Dickerson, Kemeny, Aziz, Kim, and Fahey (2004) found in an experimental study that writing about an event triggered physiological markers of shame. This study explored whether shame and guilt were triggered by perinatal death. This meant that the potential trigger for these emotions was therefore completing the demographic questionnaire and perinatal grief questionnaire which included questions about the perinatal death.

Measures and Materials

The information sheet (Appendix 3.3), consent form (Appendix 3.4) and survey were presented on the Bristol Online Survey platform. Demographic questions filtered participants into the different groups and excluded participants not eligible for the study (Appendix 3.5). Bereaved groups completed measures of grief at the beginning of the study. All participants completed other self-report measures in the order outlined below.

Perinatal Grief Scale 33 (PGS-33; Potvin, Lasker, & Toedter, 1989*)* measured grief (Appendix 3.6). This measure was used because it is widely used to quantify grief intensity after pregnancy loss (Toedter, Lasker, & Janssen (2001). It consists of three 11-item subscales: Active Grief; Difficulty Coping and Despair, each comprising 11 items. Internal reliability of the total scale is excellent, with a Cronbach's α= .94 (this sample α= .95). For the purposes of this study only the total scale was used. Only two of the PGS-33 items have guilt and/or shame emotion face validity: "I feel guilty when I think about the baby", and "I blame myself for the baby's death".

The test of self-conscious affect (TOSCA-3; Tangney, Dearing, Wagner, and Gramzow, 2000 as cited in Tangney & Dearing, 2002) measured guilt and shame-proneness (Appendix 3.7). This measure was administered in order to explore whether there were differences between guilt and shame proneness between the three groups. Tosca-3 consists of a series of brief scenarios yielding indices of shame-proneness, guilt-proneness, externalisation and detachment. The short form of this test was used and only the shame, guilt and externalisation indices were included. Internal reliability is good,

Cronbach's α = .83 for guilt-proneness and .88 for shame-proneness. Internal consistency is moderate for guilt Cronbach's α =.69 (this sample α =.62) and good for shame α =.74 (this sample α =.81). Advantages of this measure are that it measures the guilt as a reaction to the specific event in the scenario and so is consistent to the concept of guilt as a negative response to a specific action (Tangney & Dearing). It also does not require participants to distinguish between guilt and shame and is less likely to arouse a defensive reaction in some participants, as some participants may repress feelings of shame. However, a limitation is that it relies on the scenarios being the ones with which participants can identify and that they also can be seen to test moral standards.

Personal feelings questionnaire-2 (PFQ-2; Harder & Zalma, 1990) measured chronic shame-proneness and guilt-proneness (Appendix 3.8). PFQ-2 is a global adjective checklist in which participants rate the frequency they experience shame- and guilt-related adjectives. Internal consistency is acceptable, Cronbach's α= .72 for guilt (this sample =.83) and α = .78 for shame (this sample α=.87)). This measure was used because of its high face validity although a potential problem is that it requires participants to distinguish between shame and guilt (Tangney & Dearing, 2002)

State shame and guilt scale (SSGS; Marschall, Sanftner, & Tangney, 1994) was intended to measure current feelings of shame and guilt via brief phenomenological descriptions using a 5-item scale for each emotion with higher scores indicating greater feelings of shame and guilt (Appendix 3.9). The SSGS was developed to avoid the reliance on participants' ability to distinguish shame and guilt. It was used as a manipulation check for a shame induction check in an experimental study and was found to measure current feelings of shame and guilt (Tangney & Dearing, 2002). Internal consistency is good, Cronbach's α=.89 for shame (this sample α=.92) and α=.82 guilt (this sample α=.89).

Generalised Anxiety seven-item scale (GAD 7; Spitzer, Kroenke, Williams, & Lowe, 2006*)* measured anxiety across all groups. GAD-7 is a 7-item measure with excellent internal consistency, Cronbach's α= .92, this sample α = 0.91), and good test-retest reliability with a cut-off of 10 denoting moderate anxiety. Using this measure enabled a comparison of levels of distress between the bereaved and control sample.

Patient Health Questionnaire (PHQ-8; Kroenke, Spitzer, & Williams, 2001) measured depression. The 9-item version (PHQ-9) has been shown to have excellent internal consistency (Cronbach's α= .89) and good test-retest reliability (α=.84). In this study, as a condition of meeting ethics approval, the PHQ-8 was used. The PHQ-8 omits the ninth item of the PHQ-9 about thoughts of death or self-harm and has comparable operating characteristics to the PHQ-9 for diagnosing depressive orders with a cut-off of 10 denoting major depression (Kroenke & Spitzer, 2002; Kroenke et al., 2009). In this sample there was excellent internal consistency, Cronbach's α= .89. Using this measure enabled a comparison of levels of distress between the bereaved and control sample. Items 2 and 6 of the PHQ-8 items have some guilt and/or shame emotion face validity: "Feeling down, depressed, or hopeless" "Feeling bad about yourself — or that you are a failure or have let yourself or your family down." This means that it would be expected that this scale would have some correlation with the PFQ-2 and SSGS. However, other items of the measure do not have face value guilt or shame validity.

Bereaved groups were asked questions about their experience of participating in research (Appendix 3.10) and all groups were given the opportunity to comment on the survey.

Procedure

Recruitment. Recruitment of all samples was conducted online. The bereaved group were recruited through advertising websites and social media accounts of perinatal death charities and support groups. The control sample were recruited through professional and personal contacts of the researcher and advertisements on websites and local parents' pages.

Online participants. A link was provided to the online survey where the information sheet was presented. Participants were informed that while they were completing the study, they could stop at any time and their data would not be submitted. They were also informed that once they competed the survey and submitted the data, they could not withdraw from the survey because it was anonymous and it would not be possible to identify their data. Participants indicated their consent by ticking consenting statements. Participants were provided with the researcher's email if they had any queries or concerns. Completing the survey online took 35-40 minutes.

Telephone participants. Participants who participated by telephone emailed the researcher, who provided information and consent forms. Telephone participants were informed that they could stop participating at any time during the telephone call. They were informed that their surveys were not anonymous and that the researcher would inform them if their scores for GAD-7 and PHQ-8 were within the clinical range for anxiety or depression. Since their surveys were not anonymous, telephone participants were informed that they were able to withdraw from the study at any time. During the telephone call, the researcher read out the questions and completed the responses on the online survey.

Treatment of Data and Analytic Plan

Statistical analysis. A power analysis was conducted using g*power to calculate the sample size required to determine a statistical difference in state shame and state guilt between the three groups (Faul, Erdfelder, Lang, & Buchner, 2007). Since this study was speculative, there was little evidence on which to base effect sizes. However, although Schwerdtfeger and Shreffler (2009) did not report effect sizes in their comparison of women who had suffered pregnancy loss and were mothers and women who had suffered pregnancy loss who were childless, on our calculation the effect size was 0.38 (i.e. small to medium). With an alpha of 0.05 and a conservative power of 0.95, the projected sample needed was 102 for a large effect and 252 for a medium effect.

Survey responses were stored anonymously and securely, and were entered, coded and analysed using SPSS version 23. Omnibus ANOVAs, one-way ANOVAS, *t*-tests were used as appropriate in the primary analysis. In the secondary analysis, when sample sizes were small and data was not normally distributed, but distributions in each group were similar, the non-parametric tests (the Mann-Whitney test) were conducted. Pearson product-moment correlations were used to explore associations between measures amongst the bereaved sample and a partial correlation was conducted to assess whether state shame contributed to grief controlled for depression.

Missing data. There were few missing data points (0.16 % of total items). No participant had more than one missing data item from a scale or sub-scale. Three items were missing from GAD-7 and were replaced by the mean of the completed item, an approach recognised by the authors of the scale (Kroenke, Spitzer, Williams, & Löwe,

2010). Three items were missing from PGS responses, eleven from Tosca-3 responses, three from PFQ-2 and five from SSG. Each missing item was imputed as the mode score of the relevant participant, a common approach when there are only a few missing data points which has the advantage that all items are replaced by integers (Zhang, 2016).

Results

Demographic Data

Four participants elected to complete the survey by a telephone call: two in Bereaved-C and two in Bereaved-F. Demographic information for the three groups: Control, Bereaved-C and Bereaved-F is displayed in Table 3.2. Pearson's chi-square analyses were conducted to assess differences between groups. There were no significant associations between group and age, marital status and ethnicity, but there was a significant association between level of education and group χ^2 (2) =19.58, p<0.001 with the control group being more likely to be educated to degree level.

Table 3.2.

Demographic characteristics of Bereaved-C, Bereaved-F and Control groups.

Variable	*Bereaved-C* *n =40*	*Bereaved-F* *n = 55*	*Control* *n = 96*
Maternal age range (participants were not asked exact age) (median and N (%)	31-35 *n*=17 (43%)	31-35 *n*=19 (35%)	31-35 *n*=38 (40%)
N (%) Married or cohabiting with a man	35 (88%)	50 (91%)	88 (91%)
N (%) White British	4 (90%)	47 (85%)	82 (85%)
N (%) Educated to degree standard or above (%)	40 (63%)	40 (73%)	89 (93%)***
N (%) Pregnant at time of completing survey	5 (12.5%)	7 (13%)	11 (11.5%)
N (%) Partner had children from different relationship	3 (7.5%)	8 (14.5%)	9 (9.4%)
N (%) Child since bereavement	25 (62.5%)	37 (67.3%)	n/a

***denotes significant difference between groups p <.001.

Descriptive Statistics

Group mean scores for each scale are displayed in Table 3.3. To explore differences between each scale, one-way (Group x Scale) ANOVAs were performed for all scales except those measuring grief. Independent *t*-tests were conducted to explore differences between the bereaved groups in relation to the PGS. Post hoc analyses are reported by the superscripts in Table 3.3. Since women who are pregnant subsequent to experiencing perinatal death are known to have higher rates of anxiety and depression than

women who have not experienced perinatal death, a secondary analysis was conducted excluding all pregnant women from the analysis (n = 23). This data is reported in bold. Mean scores for the sub-set of Bereaved-F participants who had not a child since their bereavement (n=18) and the remaining bereaved participants (n=*77*) are displayed in Table 3.4.

Table 3. 3

Mean scores for groups for each measure and differences between groups (including scores excluding participants who reported they were pregnant)

Measure ***Excluding pregnant participants***	*Bereaved-C n=40* ***n = 35***	*Bereaved-F n=55* ***n = 48***	*Control n=96* ***n = 85***	*F*
TOSCA Shame (proneness)	25.82 *SD 6.08*	27.76 *SD 7.85*	25.10 *SD 7.43*	2.34
Excluding pregnant participants	**25.31 *SD 6.24***	**27.42 *SD 8.03***	**24.69 *SD 7.44***	**2.11**
TOSCA Guilt (proneness)	36.83 *SD 4.65*	*36.84 SD 4.11*	*36.84 SD 4.07*	0.00
Excluding pregnant women	**36.89 *SD 4.57***	**37.04 *SD 4.14***	**36.65 *SD 4.22***	**0.14**
PFQ Shame (chronic)	16.68 *SD 8.47*	17.35 *SD 7.76*	13.99 *SD 6.01*[a]	4.58**
Excluding pregnant participants	**16.86 *SD 8.95***	**17.35 *SD 7.84***	**13.46 *SD 5.25*[a]**	**5.99****
PFQ Guilt chronic)	11.85 *SD 5.70*	14.16 *SD 5.15*[b]	10.23 *SD 4.35*[a]	11.34***
Excluding pregnant participants	**11.89 *SD 5.93***	**14.08 *SD 5.16***	**10.12 *SD 4.20***	**10.19*****
SSG Shame (state)	4.63 *SD 5.34*	5.96 *SD 5.62*	2.01 *SD 3.63*[a]	13.63***
Excluding pregnant participants	**4.83 *SD 5.53***	**6.06 *SD 5.61***	**1.81 *SD 3.18*[a]**	**15.06*****
SSG Guilt (state)	5.63 *SD 5.24*	6.98 *SD 5.39*	2.59 *SD 3.71*[a]	17.59***
Excluding pregnant participants	**5.97 *SD 5.35***	**7.17 *SD 5.15***	**2.48 *SD 4.20*[a]**	**19.27*****
GAD-7 (anxiety)	10.28 *SD 6.84*	9.95 *SD 5.92*	6.13 *SD 4.45*[a]	12.63***
	10.66 *SD 7.24*	**10.03 *SD 5.68***	**6.05 *SD 4.38***	**12.87*****

Measure **Excluding pregnant participants**	*Bereaved-C n=40* ***n = 35***	*Bereaved-F n=55* ***n = 48***	*Control n=96* ***n = 85***	*F*
Excluding pregnant participants				
PHQ-8 (depression)	9.40 *SD 6.36*	10.64 *SD 6.29*	6.13 *SD 4.44*[a]	13.38***
Excluding pregnant participants	**9.49 *SD6.73***	**10.63 *SD 6.37***	**5.86 *SD 4.18*[a]**	**13.40*** **
Variables for bereaved groups				*T*
PGS (grief)	65.48 *SD 24.59*	69.78 *SD 23.73*	n/a	0.86
Excluding pregnant participants	**64.86 *SD 26.08***	**70.81 *SD 23.52***		**1.07**

*p <.05. **p<.01. ***p<.001. [a] Significant difference between Control group and bereaved groups.

[b] Significant difference between Bereaved-C group and Bereaved-F group.

Table 3. 4

Mean Scores for Subset of Bereaved-F who had not had a Child since Bereavement and Remaining Bereaved Sample

Measure	*Bereaved sample (excluding those who have not had children) n = 77*	*Subset of Bereaved-F sample who have not had children n = 18*	*p-value*
TOSCA Shame (proneness)	26.39 *SD 7.22*	29.33 *SD 6.75*	.177
TOSCA Guilt (proneness)	36.73 *SD 4.53*	*37.28 SD 3.34*	.864
PFQ Shame (chronic)	17.09 *SD 8.47*	16.94 *SD 5.99*	.75
PFQ Guilt (chronic)	12.99 *SD 5.62*	14.06 *SD 4.8*	.427
SSG Shame (state)	5.05 *SD 5.49*	6.89 *SD 5.53*	.218
SSG Guilt (state)	6.26 *SD 5.11*	7.06 *SD 6.37*	.879
GAD-7 (anxiety)	10.10 *SD 6.26*	10.06 *SD 6.62*	.943
PHQ-8 (depression)	9.66 *SD 6.23*	12.06 *SD 6.53*	.132
PGS-33 (grief)	65.94 *SD 23.52*	76.67 *SD 25.10*	.123

Hypotheses 1 and 2: State Shame and State Guilt

The relationship between state shame and state guilt for each sample was analysed in a mixed model analysis of variance (3[Control, Bereaved-F, Bereaved-C] x 2[state shame/guilt]). There was a significant main effect of group $F(2, 188) = 17.28$, $p<.001$. There was also a significant main effect of scale $F(2, 188) = 14.61$, $p<.001$. The interaction effect of group and state shame and guilt was not significant: $F(2, 188) = 0.51$, $p<.604$.

Contrasts revealed no significant difference between the Bereaved-F and Bereaved-C groups $F(1, 93) = 1.6$, $p<.209$. These data were further analysed by combining the two bereaved groups and repeating the analysis for Controls vs bereaved. In this analysis, the main effect of group was significant $F(1, 189) = 32.154$, $p<.001$, indicating that the bereaved groups experienced significantly more shame and guilt than Controls. Figure 3.1 below shows the variables fully divided by group.

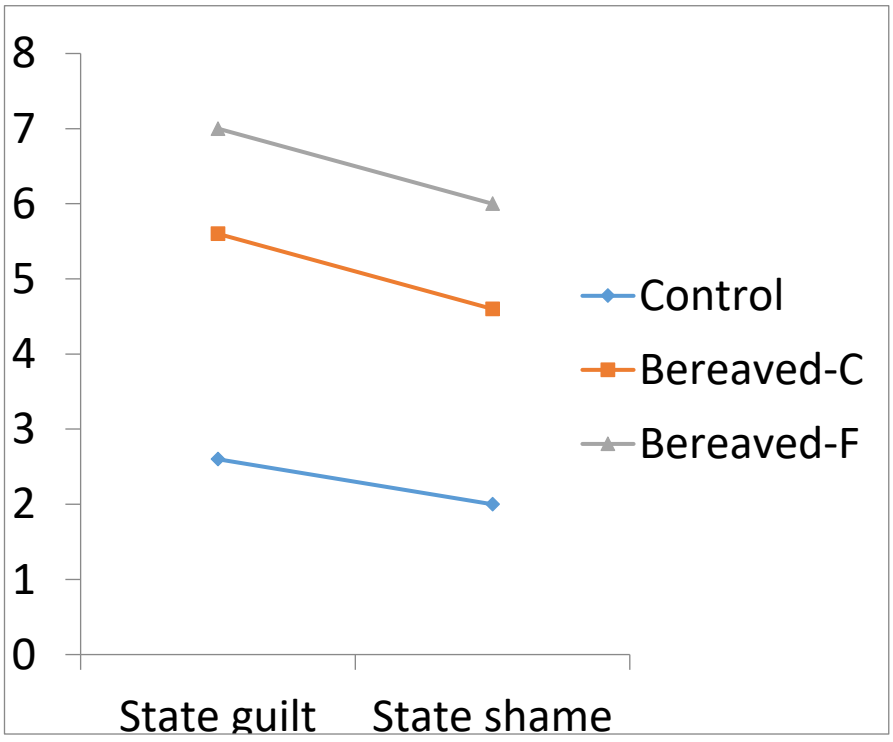

Figure 3. 1. Mean scores for state shame and state guilt in each group indicating no interaction

State shame and guilt excluding pregnant participants. The same tests were applied excluding participants who were pregnant at the time of completing the study. These indicated similar results with a significant main effect of group $F(2, 165) = 19.27$,

p<.001. There was a significant main effect of scale $F(2, 165) = 15.23$, p<.001. The interaction effect of group and state shame and guilt was not significant: $F(2, 165) = 0.47$, p<.628. Contrasts revealed no significant difference between the Bereaved-F and Bereaved-C groups $F(1, 81) = 1.15$, p<.287. These data were further analysed by combining the two bereaved groups and repeating the analysis for Controls vs bereaved. In this analysis, the main effect of group was significant $F(1, 166) = 36.71$, p<.001, indicating that the bereaved groups experienced significantly more shame and guilt than Controls.

State shame and guilt comparing subset of childless bereaved participants with remaining bereaved participants. State shame and state guilt were compared between the sub-set of the Bereaved-F group who had not had children subsequently (n=18) and the rest of the Bereaved sample who all had living children (n=77). As a result of the small sample size, it was not possible to conduct a mixed model analysis of variance so non-parametric tests were used to compare levels of state shame and guilt. A Mann-Whitney test indicated that state shame levels in Bereaved-F participants who had not had children subsequently (M = 6.89) did not differ significantly from the other Bereaved participants (M = 5.05), U = 821.50, z = 1.23, p = .218, r = .13. A post hoc power analysis revealed that a sample of 490 would be required for this difference to be significant.

A Mann-Whitney test also indicated that state guilt levels in Bereaved-F participants who had not had children (M=7.06) subsequently did not differ significantly from the rest of the Bereaved sample (M=6.26), U = 709.00, z = 0.152, p = .879.

Hypothesis 3: Group Comparisons of Guilt and Shame-Proneness and Chronic Guilt and Shame

The one-way (Group x Scale) measures of variance were not significant for either shame-proneness measured F (2,188) = 2.34, p < .1 or guilt-proneness $F(2,188) = 0.00$, p < 1.00 using the Tosca scale,.

The one-way (Group x Scale) measures of variance using the PFQ scale were significant for both chronic shame F (2, 188) = 4.58, p < .001 and chronic guilt $F(2,188)$ = 11.34, p < .001**.**

Planned contrasts revealed that compared to Controls the bereaved groups had significantly more chronic shame $t(188) = 2.88$, p <.005, r=.21 and chronic guilt $t(188)$ =

3.85, p <.001, r=.27. They also revealed that the Bereaved-F group experienced significantly more chronic guilt than the Bereaved-C group $t(188) = 2.03$, p <.045, r=.21. There was not a significant difference between the two bereaved groups in chronic shame $t(188) = .39$, p <.694.

Guilt and shame-proneness and chronic guilt and shame excluding pregnant participants. The same tests were applied excluding pregnant participants revealing similar results. The one-way (Group x Scale) measures of variance were not significant for either shame-proneness measured using the Tosca scale, F (2,165) = 2.12, p < .124 or guilt-proneness $F(2,165) = 0.14$, p < .871

The one-way (Group x Scale) measures of variance using the PFQ scale were significant for both chronic shame $F(2, 165) = 5.99$, p < .003 and chronic guilt $F(2,165) = 10.19$, p < .001**.**

Planned contrasts revealed that compared to Controls the bereaved groups had significantly more chronic shame $t(165) = 3.31$, p <.001, r=.25 and chronic guilt $t(165) = 3.71$, p <.001, r=.28. However, although the Bereaved-F group experienced more chronic guilt than the Bereaved-C group, this result was below significance $t(165) = 1.76$, p <.083, r=.19. There was also not a significant difference between the two bereaved groups in chronic shame $t(165) = .263$, p <.793.

Guilt and shame-proneness and chronic guilt and shame comparing subset of childless bereaved participants with remaining bereaved participants. Non-parametric tests were used to compare levels of shame and guilt-proneness and chronic shame and guilt between the sub-set of the Bereaved-F group who had not children subsequently and the rest of the Bereaved sample. Mann-Whitney tests indicated that, compared to other bereaved participants, Bereaved-F participants who had not had children did not differ significantly from the other Bereaved participants in relation to guilt-proneness (U = 711.00, z = 1.72, p< .864), shame proneness (U=835.00, z = 1.35, p<.177), chronic guilt (U = 726.50, z = .32, p< .75) or chronic shame (U=7.765, z = .795, p<.427).

Hypotheses 4 and 5: Group Comparisons of Anxiety and Depression between Bereaved Participants and Controls

Anxiety and depression. The one-way (Group x Scale) measures of variance were significant for both anxiety, $F(2,188) = 12.626$, $p < .001$ and depression, $F(2,188) = 13.38$, $p < .001$. Planned contrasts revealed that, compared to Controls, the bereaved groups had significantly increased anxiety, $t(188) = 5.017$, $p < .001$, $r = .343$ and depression $t(188) = 4.90$, $p < .001$, $r = .34$. Contrasts indicated that between Bereaved-C and Bereaved-F groups there was no significant difference in anxiety, $t(188) = .29$, $p = .776$ or depression $t(188) = 1.09$, $p = .277$.

Anxiety and depression excluding pregnant participants. The same tests were applied excluding data of pregnant participants revealing similar results. The one-way (Group x Scale) measures of variance were significant for both anxiety, $F(2,165) = 12.87$, $p < .001$ and depression, $F(2,165) = 13.40$, $p < .001$. Planned contrasts revealed that, compared to Controls, the bereaved groups had significantly increased anxiety, $t(165) = 4.904$, $p < .001$, $r = .343$ and depression $t(165) = 4.88$, $p < .001$, $r = .34$. Contrasts indicated that between Bereaved-C and Bereaved-F groups there was no significant difference in anxiety $t(165) = .428$, $p = .604$ or depression $t(163) = .939$, $p = .349$.

Anxiety and depression comparing subset of childless bereaved participants with remaining bereaved participants. Mann-Whitney tests indicated that, compared to other bereaved participants, Bereaved-F participants who had not had children did not differ significantly from the other Bereaved participants in relation to anxiety ($U = 685.50$, $z = -0.071$, $p = .943$) and depression ($U = 851.50$, $z = 1.508$, $p = .132$, $r = .15$). See Table 3.4 above.

Grief. An independent *t*-test conducted to compare grief revealed no significant difference between total mean grief scores $t(94) = .86$, $p = .395$, $d = 0.18$ for the Bereaved-F compared to the Bereaved-C group. See Table 3.3 above.

A g*power test conducted to calculate the sample size necessary to detect a significant difference in this sample revealed that 1,608 participants would have been required.

Grief excluding pregnant participants. An independent *t*-test conducted to compare grief revealed no significant difference between total mean grief scores $t(81) = 1.09$, $p = .280$ for the Bereaved-F compared to the Bereaved-C group. See Table 3.3 above.

Grief comparing subset of childless bereaved participants with remaining bereaved participants A Mann-Whitney test indicated that grief in Bereaved-F participants who had not had children (M=76.67) subsequently did not differ significantly from the rest of the Bereaved sample (M = 65.93), U = 855.50, z = 1.544, p = .123, r = .16. See Table 3.4 above.

Hypothesis 6: Association of Measures of Shame and Guilt with Grief

Pearson product-moment correlations were calculated for all scales in relation to the bereavement groups and are displayed in Table 3.5 below.

Table 3. 5.

Pearson Product-Moment Correlations for bereaved groups only

Scale	PGS	PHQ-8	GAD-7	State Shame (SS)	State Guilt (SG)	Tosca Shame (TS)	Tosca Guilt (TG)	PFQ-2 Shame (PFQ-S)
PGS	-							
PHQ-8	.67***	-						
GAD-7	.62***	.74***	-					
SS	.69***	.72***	.67***	-				
SG	.63***	.66***	.64***	.80***	-			
TS	.41***	.42***	,47***	.41***	.43***	-		
TG	.01	.03	.01	-.07	.05	.29***	-	
PFQ-S	.49***	.60***	,62***	.72***	.65***	.55***	.60***	-
PFQ-G	.56***	.66***	.56***	.65***	.74***	.54***	.07	.71***

****p*<.001

The Pearson product-moment correlation indicated that state shame was significantly correlated with grief r=.69, p <.01. When controlled for depression (PHQ-8) on the relationship between state shame and grief, the following partial correlation was found: r=.412, p<.001

Discussion

The primary aim of this study was to investigate differences between state shame and guilt feelings triggered by completing a demographic questionnaire which asked briefly about perinatal death and a measure of grief. It compared women who had had living children before they experienced perinatal death and those who had not. There was no significant difference between feelings of state shame and guilt between women who already had living children when they experienced perinatal death and those who had not. However, bereaved women had significantly higher state shame and state guilt than women who had had a living child between 13 and 36 months previously and had never experienced perinatal death. The secondary aim was to consider the role of shame and

guilt in grief. State shame was strongly associated with grief, explaining 48% of variability in grief and 17% of variability when controlled for depression.

Hypothesis 1: State Shame and Guilt Comparing Bereaved Participants and Controls

Consistent with the hypothesis, bereaved women had more feelings of state shame and guilt than those who had not been bereaved. While previous studies have shown an association between chronic guilt and shame and shame and guilt-proneness (Barr, 2004; Barr & Cacciatore, 2007), this study was intended to measure guilt and shame triggered by experiencing and recalling perinatal death. Although the study had no explicit reminders of the perinatal death for bereaved women, the demographic questionnaire and the perinatal grief questionnaire included questions about the death of the baby. This may have been an ecologically valid trigger reflecting the kind of trigger for shame that people who had experienced perinatal death may experience on a daily basis.

Hypothesis 2: State Shame and State Guilt Comparisons of Bereaved Participants

Contrary to the hypothesis, there were no differences between state guilt and state shame in the bereaved groups. Excluding pregnant women from the analyses did not alter these findings. Shame in perinatal death has been related to feelings of unwanted identity (Brierley-Jones et al., 2015; Ferguson et al., 2000) and this hypothesis was premised on a theory that women who already have children when they experience perinatal death may be somewhat protected from a sense of unwanted identity because their role as mother continues to be visible to the world. In this study, the majority of women had had a baby since their baby died (67% of those who had not had a child before they experienced perinatal death and 62% of those who had) and so possibly their sense of unwanted identity as someone who was not a mother had resolved. Nonetheless, comparisons of those childless bereaved women who had not had a child since their bereavement with the rest of the bereaved sample revealed that although the former group had higher shame, this difference was not significant. These results should be interpreted tentatively because of the small sample size of the childless group, but they suggest that shame may be related to other factors such as the reactions of others and disenfranchised grief in addition to a sense of unwanted identity.

Contrary to the hypothesis, the results did not reveal any differences in state guilt amongst the bereaved participants. This may reflect that women who already have

children do not have higher state guilt in relation to their families. However, possibly the measures of state guilt (SSGS) do not capture the adaptive guilt response in relation to a specific action to which Tangney and Dearing (2002) refer. Instead, they may capture a maladaptive guilt response such as self-blame which is more akin to shame. This is supported by Stroebe et al. (2014) who have shown self-blame to be associated with prolonged grief and by Kim et al. (2011) who argue that generalised guilt measures (in which he includes the guilt measures of both PFQ-2 and the SSGS) capture a free floating guilt divorced from a specific action which is equivalent to shame.

Hypothesis 3: Comparisons of guilt and shame-proneness and chronic guilt and shame

Consistent with the hypotheses, there were no significant differences between shame-proneness and guilt-proneness across the three groups. This suggests that the higher shame may have been a consequence of the experience of perinatal death rather than an unrelated factor. This tentative interpretation supports a common-sense conclusion that perinatal death had triggered feelings of anxiety and depression.

In contrast, there were differences between the bereaved groups and the control groups in relation to the chronic measures of shame. Furthermore, on the measure of chronic grief, women who had not had children previously had significantly more guilt than those who had had a child, although this result was not significant when pregnant participants were excluded from the analysis (although the effect size was similar). Since this measure was not included in the hypotheses, it is important that this result is not over-interpreted, especially because of the criticisms referred to above, that the PFQ-2 measure requires that participants have an understanding of distinctions between guilt and shame and that specifically the guilt measure may capture an emotion more equivalent to shame (Kim et al., 2011; Tangney & Dearing, 2002).

Hypothesis 4: Comparison of Anxiety and Depression between Bereaved Participants and Controls

For both bereaved groups the mean total scores for anxiety and depression were within the clinical range and had significantly higher scores than the Controls whose mean scores were within the normal range (Kroenke & Spitzer, 2002; Kroenke et al., 2009; Spitzer et al., 2006). Excluding pregnant women from the sample did not alter these

findings. This highlights that the experience of perinatal death has long-lasting effect sand is consistent with Boyle et al.'s (1996) findings that 30 months after the perinatal death, bereaved women had higher levels of depression and anxiety than non-bereaved women.

Hypothesis 5: Comparisons of Anxiety, Depression within Bereaved Participants

There were no significant differences between anxiety, depression and perinatal grief in those who had had children before experiencing perinatal death and those who had not. These findings are consistent with those of Toetder et al. (1988) and appear to conflict with Cacciatore et al.'s (2008) study which found there was a significant difference in levels of depression and anxiety. Yet Cacciatore et al.'s study had 2,292 participants and the effect was small. Within this study, the bereaved group who had not had children before bereavement had slightly higher scores for depression and perinatal grief, but not for anxiety and the power analysis indicated that with a sample of 1608 there may have been a significant effect. This highlights that having living children before experiencing perinatal death does not protect women from long-term grief. Furthermore, although mean scores were higher in relation to depression and grief, there were no significant differences in levels of depression, anxiety and grief between bereaved women with children and those who had not had a child prior to their bereavement or subsequently. Although conclusions need to be tentative because of the small sample size, this finding is consistent with Blackmore et al.'s (2011) findings that distress continues for women who have experienced perinatal death after having a living baby.

Hypothesis 6

State shame was associated with perinatal grief, having controlled for depression. This means that shame impacts upon prolonged grief independently from depression and takes Barr's (2004) understanding further, since he had highlighted that it was uncertain whether shame was still associated with grief if controlled for depression.

Limitations of this study

This study has several limitations. Only participants who could understand English could participate and so the study is limited to this population group. The survey was only advertised online and by social media and so was only available to those with devices to

access those media. However, 98.7% of the 16-44 age group have access to the internet so this is not of such a concern as previous studies (Office of National Statistics, 2016). Of more concern is that most bereaved women were recruited from support sites meaning that they may have been actively searching for support on information about stillbirths. Cacciatore et al. (2008) found that women who were actively seeking support have more symptoms than those who were not and this increases the risk of bias. Therefore, the selection effect may compromise the generalisability of the outcomes.

The study also relied on women's self-report of perinatal death without confirmation from hospital records. However, there were no inducements to participate in the survey and recruitment from NHS services was not likely to be feasible since most women would have been discharged from maternity services before becoming eligible to participate. In addition, participants were only asked to provide their age within a five year range, meaning that it was not possible to focus on age-related factors. There is some evidence that younger women may have higher levels of distress (Cacciatore et al., 2008) and so it would have been helpful to explore whether the different groups had different ages in more detail. The study also did not consider lapse of time as a factor. Nonetheless, indicators of lapse of time, such as pregnancy or having children since bereavement, were equivalent across the bereaved groups.

A further limitation of the study is that the control group participants were significantly better educated than the bereaved groups. A literature search has not found any studies stating the effect of education on shame, but lower levels of education has a small, but significant association with postnatal depression (Milgrom et al., 2008; Robertson, Grace, Wallington, & Stewart, 2004). Although this means that the Controls may have been slightly less vulnerable to anxiety and depression than the bereaved groups, this effect is small. Another grave limitation was the failure to ask women about their previous pregnancy history especially since the rationale behind the hypothesis that childless women would have a greater feeling of shame was that this shame may be rooted in a sense that their body has failed to have a child. This means that further study is required to fully explore this hypothesis.

Another limitation of the study was that a general measure of coping was not included. In part this decision was made because the measure of grief incorporated a difficulty in coping sub-measure. In part, it was made because of the necessity of being

highly selective about the measures used. A very long survey is more likely to result in participants reducing cognitive effort and moving from response accuracy to response speed (Lindell & Whitney, 2001). In addition, with an online survey there is a risk that participants will disengage with the survey before completion which would mean that their data would not be included.

A further limitation is that it was likely that there was cross-contamination of measures. Since participants' reports of their internal states were collected at the same time as the demographic questionnaire asking about their experience of stillbirth, it is possible that the correlations between stillbirth and grief, shame, guilt, anxiety and depression were inflated (Lindell & Whitney, 2001). In addition, some of the association between the shame, guilt, grief, depression and anxiety may have been an artefact of overlap in item content. This overlap seems particularly apparent in relation to the SSGS and PFQ-2 (measuring state and chronic shame and guilt respectively) and it is likely that they were measuring very similar constructs. Furthermore, there were questions both in the PGS-33 and in the PHQ-9, which have shame and guilt face validity. This means that some of the association between measures of guilt and shame and grief and depression is likely to be because these constructs are interlinked (i.e. shame and guilt are intrinsic components of grief and depression).

A final limitation with this study is that it did not use measures which captured the distinction between guilt and shame. In retrospect, to explore these distinctions properly a measure of adaptive guilt such as the Dimensions of Conscience questionnaire (Gore & Harvey, 1995) would be preferable. However, as outlined below, it may be more helpful to move away from studies focusing on these distinctions to explore how understanding these processes could develop a theoretical understanding underpinning interventions.

Clinical Significance and Further Research

The study has many strengths including the use of a control group of women of similar ages to the bereaved group and recruitment of a large sample. It is also the first study that has focused on exploring whether feelings of shame and guilt are triggered by the experience and recall of perinatal death. From a clinical perspective, it clarifies that there is no significant difference in the long-term risk of distress between women who already have living children and those whose stillborn baby is their first child. This study

also provides some evidence that mothers continue to grieve after subsequently giving birth to a healthy baby subsequently since there was no significant difference between childless bereaved women who had not given birth subsequently and bereaved women who had a child at the time of taking the survey. However, further research into this area is important since the sample size of women who had not had a baby previously and had not given birth subsequently was small. Furthermore, further research focusing on the association of experiences of miscarriages and fertility interventions feelings of shame and guilt in perinatal death is necessary to explore the hypothesis that shame in perinatal grief is rooted in a denial of identity as a mother.

A strength of the research into perinatal bereavement is that researchers come from several different professional fields including obstetrics, nursing, midwifery, social work and psychology. However, this may result in a tendency for the body of work to be atheoretical. Both the present and previous studies on shame and guilt following perinatal bereavement may be limited by not drawing on the most contemporary of theoretical models of self-conscious emotion. Indeed, a recent Cochrane review in relation to support for mothers, fathers and families after perinatal death, Koopmans, Cacciatore and Flenady (2013) did not find any intervention to review. Such research is vital given the long-term consequences of perinatal death for many women.

Stigma and disenfranchised grief are frequent themes in the literature relating to perinatal grief (Brierley-Jones et al., 2015; Burden et al., 2016; Campbell-Jackson, Bezance, & Horsch, 2014). Such feelings could be a key element in shame and go further than the concept of unwanted identity which underpinned the hypotheses in this study. Further research on this field could focus on the operation of this shame within a theoretical framework relating to this concept. Gilbert's work on shame may be an appropriate model (Gilbert, 2003). He builds on Lewis' work, but emphasises the role of people's relationships with others, arguing that we feel shame when we believe that we are not meeting others' standards of ourselves (as cited in Tangney & Dearing, 2002).

This conceptualisation of guilt and shame has been the springboard to Gilbert's development of compassion focused therapy (CFT) which is particularly targeted at people with high levels of self-criticism and shame. CFT is a trans-diagnostic therapy rooted in evolutionary and social psychology, attachment theory, neuroscience and cognitive-

behavioural therapy (Gilbert, 2014). The main focus is to encourage emotional and mental wellbeing through developing compassion towards self and others. The therapy has been used within several clinical populations including eating disorders, psychosis and perinatal distress (Braehler et al., 2013; Cree, 2010; Gilbert, 2009; Goss & Allan, 2010). Although further studies are required, an early systematic review found that initial results are promising (Leaviss & Uttley, 2015). Hence, given the role of shame in prolonged grief, CFT may be an appropriate therapy for people who have experienced perinatal death.

Conclusion

This study has found that there are no differences in levels of shame and grief between women who experience perinatal death when they have living children and those who have no children when they experience perinatal death. It provides evidence that 13-36 months after the perinatal death, women have higher levels of distress than women of similar ages who have had living babies and no experience of perinatal death. It also suggests that shame plays a key role in the long-term grieving of women who have experienced perinatal death. Future research could focus on drawing on theoretical models of self-conscious emotions to develop interventions targeted at dealing with shame such as compassion-focused therapy.

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Executive Summary of Main Research Project

Almost 1% of births in England and Wales resulted in perinatal death defined as stillbirth at more than 24 weeks of gestation or neonatal death of a baby within 28 days of birth. Yet perinatal death is often viewed differently from other bereavements with qualitative studies indicating that women find that their grief is minimised by friends, relatives and healthcare professionals who view a perinatal death as a less traumatic death than that of a partner or older child. Although most women who experience a perinatal death recover within 12 months of the loss without professional support, 15 to 25% continue to experience clinically significant symptoms 12 months later.

Several studies have investigated factors associated with poorer outcomes for women bereaved through perinatal death. Higher levels of distress are associated with poor levels of social support, more recent loss and pre-existing mental health problems. Research into the impact of having living children before experiencing perinatal death has had mixed findings with some studies finding that they have lower levels of distress and others finding no difference between them and those who have not had a child. This means that no firm conclusions can be drawn about whether having a child before experiencing perinatal death is associated with higher or lower levels of long-term distress.

The role of guilt and shame in prolonged grief reactions in perinatal death has been explored in some empirical studies. The words shame and guilt are sometimes used interchangeably, but within psychology guilt and shame often describe slightly different feelings. Shame is an emotion whereby yourself, not just your behaviour is negatively evaluated. It is often accompanied by a sense of wanting to hide flaws and worthlessness and powerlessness. Guilt is an emotion in which behaviour is negatively evaluated, but not the whole self.

In a longitudinal study, Barr (2004) studied the association between guilt and shame-proneness and perinatal grief for men and women at one month and then thirteen months after the death of a baby. He found that shame-proneness, but not guilt-proneness explained a small variance a month after the stillbirth, but that 13 months later, shame-proneness explained a substantial proportion of the variance in women's late grief. This means that further investigation into why women who have experienced perinatal death

may experience such feelings and what predicts these feelings is important to a better understanding of prolonged grief.

One line of inquiry is to explore differences in feelings of guilt and shame between women who already had living children and those who had had no children at the time of the perinatal death. In particular, a qualitative study found that some mothers reported denial of their identity as a mother as a result of stillbirth. A comparison of infertile couples with couples who had a healthy child, indicated that infertile couples had significantly more feelings of shame. In contrast, Avelin (2011) suggests that where mothers have surviving children, they can feel guilty in relation to their family because of not being able to do things that they feel they should do for their children because of their grief. From this evidence, it was hypothesised that a risk factor for state shame in perinatal loss may be to have never had a living child, while a risk factor for state guilt may be having surviving children.

The research study was therefore in part to investigate whether feelings of state shame and state guilt between 13-36 months after perinatal death are predicted by whether a women has had living children prior to their perinatal loss. It also extended Barr's (2004) study in considering whether state shame and guilt triggered by recalling experiences of perinatal death is associated with grief controlled for depression.

Three groups of adult female participants were compared: 55 women in the bereaved first (Bereaved-F) sample had not given birth to a living child before the death of their baby; 40 women in the bereaved group with children (Bereaved-C) sample had a living child born before the perinatal death; and 96 women in the control sample (Control) had had a living baby between 13 months and three years previously and had never experienced perinatal death of a baby. The two bereaved groups completed a questionnaire on grief and all groups completed measures of guilt and shame-proneness, chronic guilt and shame, state guilt and shame and anxiety and depression.

There were no differences between feelings of state shame and guilt between women who already had living children when they experienced perinatal death and those who did not. However, bereaved women had significantly higher levels of state shame and state guilt than women who had had a living child between 13 and 36 months previously and had never experienced perinatal death. However, state shame was strongly associated

with grief explaining 48% of variability in grief and 17% of variability when controlled for depression.

This study provides evidence that 13-36 months after the perinatal death, women continue to have higher levels of distress than women of similar ages who have had living children and no experience of perinatal death. It also suggests that shame plays a key role in the long-term grieving of women who have experience perinatal death. Future research could focus on drawing on theoretical models of self-conscious emotions to develop interventions targeted at dealing with shame such as compassion-focused therapy.

Connecting Narrative

As I reviewed the case studies and research studies comprising the portfolio, I became aware of several recurring themes including the therapeutic role of formulation, a focus on blocks and barriers to therapy both in relation to individuals and an interest in bereavement and grief. The first two themes shaped my case studies.

Case Studies

In my first case study, I investigated the role of formulation in treatment for a woman with severe anxiety who had been abused by her father. I found in this early piece of work that formulation was helpful, but active interventions based on the formulation were also important to creating change. My second case study explored whether a neuropsychological assessment could be therapeutic for a man who had had a stroke and not come to terms with his difficulties. Formulating and providing a better understanding of his problems was important to my client's adjustment to loss since it enabled him to make changes. However, he also had to work hard to develop new ways of living which incorporated this understanding. Through this focus on formulation, I have developed a better understanding of its role and value, but also greater awareness that interventions are also necessary for change.

The importance of addressing barriers and blocks to therapy was a significant theme across three of my case studies. In my third case study, I was stuck in my work with a boy with a restricted diet, partly because his mother was focused on finding a diagnosis for his problems. Diagnoses had been explored by other professionals, but they had believed anxiety to be the root of his problems. Prompted by my supervisor, I had a session with his mother alone where we discussed her feelings and understandings about what anxiety means and acknowledged her difficulties. It was only through addressing her fears that we were able to move on with therapy.

Barriers to engagement in therapy arose in a different context in my fourth case study when I developed a mindfulness group for people with learning disabilities. In preparing therapeutic sessions, we had to consider how people could physically get to sessions, the benefits and drawbacks to case workers attending and then think carefully about creating useful and accessible mindfulness exercises. During my fifth case study, I thought more about barriers to therapy because my fifteen year old client was in kinship

care and had a reputation for being difficult to engage. In addition to focusing on practical issues such as location of therapy and providing a safe space, I explored how cognitive behavioural therapy focused on a current problem could be used to develop trust and therapeutic alliance so that a looked after child could begin to think about how some of her early experiences may have affected her. Through these cases, I have learned that exploring what prevents people from accessing, attending or making use of therapy is fundamental to achieving change.

Research Projects

Before I began training, my clinical experience was with primary care adult mental health and CRUSE bereavement care and my continuing interests in adult mental health and grief are reflected in my three research projects.

Service Improvement Project

In my Service Improvement Project, I explored how Recovery Navigators (RNs) and Recovery Practitioners (RPs) make decisions to refer people for psychological input within Recovery and Assessment team (RS) which is equivalent to a community mental health team. Falguni Nathwani (FN), a clinical psychologist and course supervisor, who works for complex psychological interventions (CPI), presented this idea at the research fair.

At this stage, we had no knowledge at all about the decision-making process of RPs and RNs. Therefore, we chose a qualitative design because we wanted to provide rich contextual information to CPI about how RNs and RPs made decisions to refer and a survey would not have provided this information. Once the project was designed, obtaining ethical was a relatively straightforward process. The University Ethics Committee approved the idea quickly and the NHS Research & Development officer approved the study as an evaluation project.

The original intention had been to hold two focus groups, one with recovery navigators, the second with community psychiatric nurses. After a team meeting in which I presented the project and invited people to participate, the team manager spoke passionately about her belief that social workers and occupational therapists who are recovery practitioners should not be excluded from the study since they also made

referrals. As a result of this discussion, we expanded the criteria for participants so that we recruited for RPs from any profession.

Although I opened up the research to all RPs, different people work in different shift patterns and I also had limited flexibility about the hours I could interview which meant I found it impossible to arrange focus groups. Therefore, I interviewed people individually and in groups trying to ensure that I had asked everybody the same questions, but at the same time allowed people space to comment on additional concerns.

I did not know any of the people who I interviewed and believe this may have enabled participants to be very honest. KB, who coded the interviews, commented that she was surprised at how frank people seemed to have been able to be. Nonetheless, I believe that people may have had different responses depending on their interview grouping. It was noticeable that the two individual interviewees were more expansive than others about their lack of knowledge about the referral criteria and I have wondered whether this reflected their individual experience or whether they felt more able to talk more freely.

I enjoyed the conducting the analysis of the interviews. I was fortunate that my psychology degree at the Open University emphasises the importance of qualitative research and so I had some previous experience of thematic analysis. In addition, Vuokko Wallace gave me helpful advice when I was having difficulty working at a consistent structure while KB was a constant source of support and advice.

The process of feeding back at the CPI meeting was interesting. I knew many of the CPI professionals because my first placement had been in that team and this made giving the talk easier. It was helpful to have their views and comments on the material before providing final recommendations for change as I was able to incorporate their feedback especially in relation to experiences of training. However, it was only at this late stage that I realised that two other people in my cohort had also done projects within the RS. I wish that I had worked in a more joined-up way with them earlier and wonder if the course could advise each cohort to liaise closely with each other about their projects.

Critical Review of the Literature

As a trainee with limited research experience, I found identifying a topic for the critical review of the literature difficult. However, I was interested in researching the

effects of bereavement and a conversation with a professional from the child oncology unit lead me to sibling bereavement. I selected a systematic review to bring a level of methodological rigour especially because this was an area in which the quality of studies was notoriously poor. This involved a review protocol which pre-defined each of the processes involved including the terms used to search for papers, search strategy, identification of papers for inclusion, data extraction and assessment of study quality. In conducting a systematic literature review, it is important to include more than one reviewer. I worked with a fellow trainee who reviewed 20% of the abstracts and titles and then a further 20% of the full papers. Ideally, she would have reviewed all the papers, however, time constraints meant that this was impossible.

I found the review process difficult especially because there was not a specific quality assessment tool suitable for every study and there were several different designs and outcomes measured. However, the experience influenced my main project leading me to think more carefully about the importance of clear hypotheses and analysis plan. My research tutor guided my critical thinking about the literature and understanding of results, but I found structuring the review itself difficult and perhaps could have found more support helpful. Initially, I had simply summarised the results on a study by study basis and it was only by immersing myself in the literature in this way that I felt confident to synthesise the results by outcome. Once I did so, thinking about the studies in the context of Stroebe et al.'s framework highlighted how few studies have focused on the processes involved in maintaining and recovering from grief and that many of the studies do not have a theoretical basis. I also became aware of how family dynamics often seemed central to the psychosocial outcomes for bereaved siblings. This central role of a child's family relationships was a recurring theme also within both my CAMHS placement and my placement with Thinking Allowed.

Main research project

I first became aware of stillbirth and neonatal death sixteen years ago when I was pregnant with my oldest child. My sister was working as a doctor in the Gambia at the time and she was acutely aware of the risks of childbirth for both child and mother. I became much more conscious that stillbirth is still common in developing countries and that it continues to happen in the UK. When I had my children, I was acutely aware of this risk. I have not had experienced perinatal death, but women who have this experience

have lived some of my deepest fears. When Sarah Stacey who was working in the specialist Neonatal Unit at St Michael's Hospital said she would be interested in supervising a project in women bereaved through perinatal loss, I was keen to investigate the psychological processes in grief and explore how this could be clinically helpful.

The idea of comparing women's experiences arose from a conversation with Sarah in which she talked about her clinical experience that women who had children seemed to feel more guilt than shame while women who had no children seemed to feel more shame than guilt. In the initial stages of developing the project, I contacted Bristol Stillbirth and Neonatal Death Charity (Sands) to ask if they would meet up to discuss my tentative ideas. They put me in touch with Veronica Lee, a retired chaplain who had worked at Southmead Hospital with a special interest in perinatal death. She was interested in exploring the ideas of shame and guilt and focused on the idea that some women felt shame that their body had not been able to have a baby while others felt guilty for trying. I learned more about this distinction between guilt and shame through discussions with Megan Wilkinson-Tough, my internal supervisor who helpfully guided me towards the literature.

As a result, I developed a proposal to look at this difference. I thought carefully about how we could have a control group to compare their feelings of shame and guilt with people who had experienced childbirth and eventually decided this should be women of the same age who had had children within the same frame. We knew that this group may have increased rates of anxiety and low mood, but we also believed that they would be of similar age and experiences.

Once I had a clearer idea of the project, I consulted with two representatives from Sands in Bristol. They were concerned about the time period I was focused on because I was hoping to recruit women who had experienced perinatal death between 7-18 months after the perinatal death. They commented that at the time of the first anniversary of the baby's death it was particularly difficult and that in particular the questionnaires about shame and guilt-proneness which included scenario based questions were likely to seem irrelevant. Both reflected that they believed that mothers who already had children had different experiences of grieving than those who did not have children. They expressed a preference for an online survey, saying that this felt more private and convenient than a survey by phone or face to face. Their views shaped my application for NHS ethics.

My application to NHS ethics was a bruising experience. I had checked carefully that they had telephone conference technology prior to booking. However, this was not available and so the Research and Ethics Committee was conducted by mobile and it was unclear if all committee members could hear what was said. A midwife who had conducted some research into early grief responses led the process. She was adamant that an online survey was inappropriate to investigate grief. I felt very frustrated that my consultation with people with personal experience and research in relation to people's experiences of participating in surveys was dismissed (Sveen, Eilegard, Steineck, & Kreicbergs, 2014). In addition, I was not given opportunity to address their concerns about some of the issues. For example, the committee were concerned about use of PHQ-9 because of the last question which asks about suicide, but did not allow us to explore using the PHQ-8 because they were unaware that this was also validated. Since this experience, I have read an article by Della Sala and Cubelli (2016) in the Psychologist which conveys many of my frustrations of the experience.

Once I had received an unfavourable opinion, I decided to advertise the survey online through bereavement charities. Although the NHS experience was difficult, I learned a lot from this experience, helped by the support of internal and external supervisors and my clinical tutor. In my application for University ethics, I was careful to address the concerns that the NHS committee had raised and believe my project was more robust as a result of doing so. I took the following steps:

- I consulted with Becca Swingler, a friend who is a consultant obstetrician who had worked with parents frequently after they had experienced perinatal death and Fiona Green, a midwife who had worked closely with Sands to discuss the project. They both advised that they thought that guilt was often experienced by people who had experienced perinatal death.
- I redesigned the study so that participants could opt to participate by telephone if they preferred.
- I removed qualitative questions and references to shame and guilt.
- I added questions about the research experience modelled on Sveen et al. (2014) which asked about siblings' experience of participating in research.

- I piloted the survey with committee members from Bristol Sands which raised important issues of how people would feel on exclusion which were very relevant when the project went live.
- I also attended a Sands conference where I networked and discussed research with people. This was important as it enabled people to put face to name and the person I met there became my contact throughout.

Sands asked me to make further changes. In particular, they asked me to change wording I had included before the questions on state shame and guilt where I stated "When you think about your baby." We were concerned that this change may mean that state shame and guilt might not be triggered although this concern was not realised in our results. I enjoyed the collaboration with Sands. I was meticulous about responding to their comments and their input was invaluable on issues such as payment for participation and avoiding the use of "loss" instead of death. However, they listened when I explained why I wanted to use the perinatal grief scale which contains a question about considering suicide and agreed to my doing so. Working with Sands so they would advertise the research gave me valuable lessons in the importance of working with people who know a field well and listening carefully to their advice.

When the research eventually went live, I was amazed by how many women contacted me who wished to be involved and saying that by doing so they felt they were doing something positive and remembering their baby. Through this experience, I also learned about some of the controversies surrounding perinatal death such as the cut-off time for a baby being given a birth certificate and some men's sense that they have no voice when their baby died. Both women and men contacted me to ask questions about how I reached my exclusion criteria and I had to think very carefully about how I responded, realising how sensitive and difficult these issues are. Thus, people who were bereaved tended to want to speak more about their experiences and their baby. In contrast, several friends felt uncomfortable about sharing research because they had friends who were pregnant. These experiences lead to thinking deeply about some of the social issues around perinatal death. I became aware of how seldom women feel able to talk about what has happened to them and even say their babies' names. It highlighted to me what a difficult and lonely experience women bereaved through perinatal death can have.

I feel passionately about the people I have worked with especially in my main project and case studies. It has sometimes been very hard work and overwhelming, but I wonder whether this reflects the reality of completing research when not a trainee too! I would love to work in this field again and have many ideas which I would like to develop particularly in relation to stigma and social relationships in women who have recently had children. I regret that I have not yet published any of my work. While I have been doing this course, this extra step has seemed one hurdle too many. However, I am proud to have been accepted to give both an oral presentation of my main research project at an international bereavement conference. I hope to publish the research studies in this portfolio as well as writing a paper on bereaved women's participation in research.

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To Ed, Tom, Martha and Esther. There are not enough words in the world to say how lucky I am to have you and how much I love you.

In memory of

My mother, Susan Mary Hamer, who would have loved this

and

Emma Louise Rees

Appendix 1

Appendix 1.1. Journal information

OMEGA – Journal of Death and Dying – 5 year impact rating 0.851: Instructions to Authors

Instructions for Authors

Manuscripts can be sent in APA style to **https://mc.manuscriptcentral.com/omega.**

Please refer to the latest Publication Manual of the American Psychological Association. A synopsis of this manual is available from the American Psychological Association. http://apa.org/

Originality Authors should note that only original articles are accepted for publication. Submission of a manuscript stands for certification on the part of the author(s) that neither the article given, nor a version of it has been published, or is being considered for publication elsewhere.

Format Prepare manuscripts according to the latest Publication Manual of the American Psychological Association. A synopsis of this manual is available from the American Psychological Association. http://apa.org

Manuscripts Manuscript must be word processed, double-spaced, with wide margins. Paginate consecutively starting with the title page, which should be uploaded as a separate file. The organization of the paper should be shown by right headings and subheadings. Please be sure to remove all self-identifying information from the manuscript file before sending. Author information should only be included on the title page.

Style Technical terms specific to a particular discipline should be defined. Write for clear comprehension by readers from a broad spectrum of scholarly and professional backgrounds. Avoid acronyms and footnoting, except for acknowledgments.

Permissions Authors are responsible for all statements made in their manuscript and for obtaining from copyright owners to reprint or adapt a table or figures, or to reprint a quotation of 500 words or more. Authors should write to original author(s) and publisher to request nonexclusive world rights in all languages to use the material in the article and in future editions. Provide copies of all permission and credit lines obtained at the time of manuscript submission.

Manuscript Submission Guidelines:

Manuscript must be word processed using Word or Open Office Writer, double-spaced, with wide margins. Paginate consecutively, starting with the title page.

Title Pages should be uploaded as a separate file and include the follow as is applicable:

• Full article title

• Acknowledgements/credits

• Each author's complete name and institutional affiliation(s)

• Grant numbers and/or funding information

• Corresponding author (name, address, phone/fax, e-mail)

• Up to five keywords as it should appear if it were to be published.

Abstracts of 100 to 150 words are required to introduce each article.

Manuscripts should be saved in a Word .doc or .docx file type. The organization of the paper should be indicated by appropriate headings and subheadings.

Please be sure to remove all self-identifying information from the manuscript file before submitting.

When possible, all illustrations, figures, and tables are placed within the text at the appropriate points, rather than at the end. If this is not possible:

Figures should be referenced in text and appear in numerical sequence starting with Figure 1. Line art must be original "drawings" in black ink proportionate to our page size. Indicate top and bottom of figure where confusion may exist. Labeling should be 8 point type. Clearly identify all figures. Large figures should be drawn on separate pages and their placement within the text indicated by inserting:

Insert Figure 1 here

Tables must be cited in text in numerical sequence starting with Table 1. Each table must have a descriptive title. Any footnotes to tables are indicated by superior lower case letters. Large tables should be typed on separate pages and their approximate placement indicated within text by inserting:

Insert Table 1 here

Appendix 1.2. Critical Review of literature – Search information

Search conducted on 18 August 2016

Apa psych info – using index terms ({siblings} OR {brothers} OR {sisters}) AND ({bereavement} OR {grief}

Results = 240 initially; 133 in journals.

Pub Med:

Results =255 initially; 236 when reduced to English language

Scopus: TITLE-ABS-KEY ((bereavement OR grief)) AND TITLE-ABS-KEY ((siblings OR brothers OR sisters))

= 706 articles

Limited to articles and reviews

= 624

Limited to English language

= 565

World of Science; **TOPIC :**(bereavement OR grief) *AND* **TOPIC :**(siblings OR brothers OR sisters)

Timespan: All years. **Indexes:** SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC.

=291 articles

Refined to articles and reviews and English language – 258 articles

258

+565

+236

+133

=1,192

693 studies after all duplicates removed

Appendix 1.3 Critical Review of the Literature – validated instruments assessing psychosocial adjustment

Alphabetised list of validated instruments assessing psychosocial adjustment.

Assessment tool	*What it tests and validity and reliability data*	*Studies using instrument*
1.Adult Self-Report (ASR) **(Achenbach & Rescorla, 2003)**	**Emotional and Behavioural difficulties:** List of questions asking how often the participant has had a list of emotional or behavioural problems in the last six months. The scale splits into internalising and externalising scores. Internal reliability .89 for internalising score and .93 for externalising score.	Stikkelbroek, Bodden, Reitz, Vollebergh, and van Baar (2016)
2.Best friends nomination (BFN) **(Bukowski & Hoza, 1989)**	**Social relationship:** Students nominate three best friends from a list of classmates to indicate a social preference score for total number of nominations and mutual friendship. A literature search was undertaken, but it was not possible to find data in relation to reliability and validity.	Gerhardt et al. (2012)
3. Children's Behaviour Checklist (CBCL) **(Achenbach & Edelbrock, 1983)**	**Emotional and Behavioural difficulites:** Standardised norm referenced instrument that provides clinical data. Parent's form has two scales: the Total Behavour Problem (comprising the Internalising Behaviour Problem Scale and Externalising Behaviour Problem Scale) and the Social Competence Scale. The teacher's form has the same Total Behaviour Problem Scale and also an Adaptive Functioning Scale. A referable score is defined as any Total Behaviour Problem score in the 90th percentile in the normative sample. Reliability= .94-.96 for parent form and .90 to .99 for teacher form (Achenbach & Edelbrock, 1983).	Birenbaum, Robinson, Phillips, Stewart, and McCown (1989); Davies (1988) Demi and Gilbert (1987); McCown and Pratt (1985)
4. Child Depression Inventory (CDI) **(Kovacs & Beck, 1977)**	**Depression:** A 27 self-report scale for children aged 7-17 years. Internal consistency reported at .86.	Demi and Gilbert (1987) Morris et al. (2016)

Assessment tool	*What it tests and validity and reliability data*	*Studies using instrument*
5. Child reaction index (CRI) **(Frederick, 1985)**	**Trauma:** 20-item questionnaire administered in interview format to inquire about child's reactions to traumatic event. Frederick reports a .91 correlation between CRI scores and established cases of PTSD.	Applebaum and Burns (1991)
6. Hospital Anxiety and Depression Scale (HADS) **(Zigmond & Snaith, 1983)**	**Anxiety and depression:** A self-report scale with seven items each and providing an anxiety scale, a depression scale and a total score scale. The reliabilities were .82, .77 and .86 respectively (Crawford, Henry, Crombie, & Taylor, 2001).	Eilegard et al. (2013b)
7. Hopkins Symptom Checklist (HSCL) **(Derogatis, Lipman, Rickels, Uhlenhuth, & Covi, 1974)**	**Somatisation, depression, anxiety, interpersonal sensitivity and obsessive compulsiveness:** A 58-item self-report symptom inventory. Reported test-retest reliability .75 to .82 and internal consistency .84 to .87.	Demi and Gilbert (1987)
8. Inventory of Complicated Grief Questionnaire (ICG) **(Prigerson et al., 1995)**	**Complicated grief:** A 19 item self-report scale. Internal consistency reported at .9 and test-retest reliability .8.	Sveen, Eilegard, Steineck, and Kreicbergs (2014)
9. Impact of Event Scale (IES) **(Horowitz, Wilner, & Alvarez, 1979)**	**Trauma:** A 15 item self-report scale. Internal consistency reported to be .79 to 92.	Demi and Gilbert (1987)
10. Kessler-6 Psychological Distress Scale (Kessler et al., 2003)	**Anxiety and depression:** A six-item scale screening. Internal reliability .89-.92.	Rosenberg et al. (2015)
11. Louisville Behavior Checklist (LBC) **(Miller, 1984)**	**Emotional and behavioural difficulties:** A 164-item inventory of childhood adjustment completed by parents with standardised norms.	Mulhern et al. (1983)

Assessment tool	*What it tests and validity and reliability data*	*Studies using instrument*
12. MOS social support survey (Sherbourne & Stewart, 1991)	**Social support:** A nine-item survey assessing functional dimensions of general social support, including emotional, informational and interactive domains. Total scores are summed and then transformed to a 100 point scale with higher scores indicating greater social support. The average social support score among US adults is 70.1.	Rosenberg et al. (2015)
13. Self-image questionnaire (OSIQ) **(Offer, Ostrov, & Howard, 1977)**	**Self-concept:** 11 dimensions of self-concept: impulse control, emotional tone, body and self image, social relationships, moral values, sexual attitudes, family relationships, mastery of external world, vocational educational goals, psychopathology and ego strength. By summing scores a total OSIQ score is found. The mean on the OSIQ is 50 and the standard deviation is 15 for adolescents of the same-sex, same race and like age group. Scores above 50 indicate better adjustment than demonstrated by normal youths in their norm group. Good internal consistency = .6 for 8/10 scales (sexual attitudes was not tested)(Patton & Noller, 1994).	Balk (1983a, 1983b, 1990, 1991)
14. Piers-Harris Self-concept Scale (PHSCS) **(Piers, 1969)**	**Self-concept:** A self-report instrument designed for children aged 8 to 18 years consisting of 80 first person statements requiring a yes/no response. The scale was standardised on 1,183 children in grades 3 to 12 in a school district in Pennsylvania. The Kuder-Richardson co efficients range from .77 to .78 and the scale is considered to have good internal consistency and adequate temporal stability.	Martinson, Davies, and McClowry (1987)
15. Child version of Los Angeles PTSD Reaction Index (PTSD-Ri) **(Rodriguez, Steinberg, & Pynoos, 1999)**	**Trauma:** Self-report questionnaire assessing PTSD according to DSM-IV criteria with an internal consistency of .89.	Morris et al. (2016)

Assessment tool	*What it tests and validity and reliability data*	*Studies using instrument*
16. Revised Class Play (RCP) **(Masten, Morison, & Pellegrini, 1985)**	**Social relationships:** A descriptive matching instrument which asks students and teachers to imagine that they are the director of a play and to cast members of the class into 42 hypothetical roles. Students could be picked to play more than one role. Item scores reflecting teacher selections or the number of peer nominations each child received for each role are created. Shows good internal consistency =.81-.95 across age groups.	Gerhardt et al. (2012)
17. Rutter Child Scale (RCS) **(Rutter, Tizard, & Whitmore, 1970)**	**Behavioural problems:** Behaviour checklist which consists of 26 descriptions of various behaviours which are rated on a three-point scale from 0= doesn't apply, to 1= applies somewhat and to 2= certainly applies. Rutter reported a test-retest coefficient of .89 after two months.	Pettle Michael and Lansdown (1986)
18. Self-concept Scale (SCS) **(Lipsitt, 1958)**	**Self-concept:** A paper and pencil test in which children rated themselves in response to a list of characteristics as they are or would like to be. The discrepancy between the two is considered a measure of the children's satisfaction with themselves. A third scale "My sister/brother as he was" is added as a measure of the child's view of the sibling. It has not been possible to find validity and reliability data for this measure.	Pettle Michael and Lansdown (1986)
19. Youth Self-report (YSR) **(Achenbach, 1991)**	**Emotional and Behavioural problems:** List of questions asking how often the participant has had a list of emotional or behavioural problems in the last six months. The scale splits into internalising and externalising scores. Internal reliability .85 for internalising score and .87 for externalising score.	Stikkelbroek et al. (2016)

Appendix 1.4 Critical Review of the Literature – validated instruments measuring factors associated with psychosocial outcomes

Alphabetised list of validated instruments measuring factors associated with psychosocial outcomes.

Assessment tool	*What it tests and validity and reliability data*	*Studies using*
1. Alabama Parenting Questionnaire (APQ) **(Scott, Briskman, & Dadds, 2011)**	**Parenting behaviours:** Self-report measure with four subscales (positive parenting, inconsistent discipline, corporal punishment and parental involvement) on a five point scale. Internal consistency for each study ranged between .72-.9.	Morris et al. (2016)
2. Family Environment Scale (FES) **(Moos, Insel, & Humphrey, 1974)**	**Family functioning:** A 90-item standardisd questionnaire which results in scores indicative of family functioning.	Mulhern, Lauer, and Hoffmann (1983)
3. Minesota Multiphasic Personality Inventory - 138 (MMPI-138) **(Overall, Higgins, & de Schweinitz, 1976)**	**Parents psychopathology and personality:** An abbreviated version of the Minnesota Multiphasic Personality Inventory, a widely used measure of psythopathology and personality which results in standard scores and clinical scales as the long form.	Mulhern et al. (1983)
4. Prolonged Grief Disorder Questionnaire (PG-13) **(Prigerson et al., 2009)**	**Parents' grief:** PG-13 is composed of 12 grief symptoms answered on a five-point Likert Scale. Total score indicates internal consistency of .93.	Morris et al. (2016)

Assessment tool	*What it tests and validity and reliability data*	*Studies using*
5. Parental Role Scale (PRS) **(Weissman & Bothwell, 1976)**	**Parental role:** A subscale of the Social Adjustment Scale Self-report (Weissman & Bothwell, 1976) with internal consistency reported as .76.	Demi and Gilbert (1987)
6. PTSD Checklist (PTSD CL) **(Weathers, Litz, Herman, Huska, & Keane, 1993)**	**Parents' trauma:** self-report questionnaire consisting of 17 questions relating to post-traumatic stress disorder (PTSD) according to the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV) criteria (APA, 2000). It has an internal consistency of .95.	Morris et al. (2016)

Appendix 1.5.

Critical review of the literature. Reasoning for quality evaluation.

Study and number of participants	Selection bias	Control group	Associative factors (bias where retrospective reporting or not self-report)	Confounds	Outcomes	Statistics	Quality Total
1. Applebaum and Burns (1991) (n=20)	Small sample, selection from self-help groups: **0**	No control group: **0**	PTSD of parents – tested at time, but not validated questionnaires: **1**	Siblings from same family not included. No testing of PTSD prior to Index Child death**: 1**	Outcomes for siblings tested at with self-report validated questionnaires and observer interviews: **2**	Statistics described. Hypothesis clearly reported**: 2**	**6**
2. Balk (1983a); Balk (1983b) (n=33)	Small sample, selection from self-help groups: **0**	No control group: **0**	Age of adolescents Change of grief reactions over time retrospective self-report: **1**	Siblings from same family not included, but no pretesting: **1**	Outcomes tested with validated self-report measures: **1**	Statistics described. Hypotheses not clearly reported: **1**	**4**
3. Birenbaum, Robinson, Phillips, Stewart and McCown (1989) (n=61)	Small sample, selection from medical centres: **1**	No control group: **0**	Changes over time (measured at 4 different collections points) –but sample very small: **1**	Pretesting (but not before illness) Siblings accounted for: **1**	Outcomes tested with validated questionnaires from parents and teachers: **2**	Statistics described. Hypotheses not clearly reported: **1**	**6**
4. Bolton et al. (2016)(n=7243)	Large population-based cohort study: **2**	Control group non-bereaved siblings matched on age, gender and income: **2**	Sibling as only option Sibling gender Marital status Age of Index child at time of death. Age of sibling at age of death: **2**	Pretesting (2 years prior to death). All siblings in each family included and all types of death included: **2**	Outcomes tested on hospital discharge abstracts and physician billing records: **2**	Statistics described. Hypothesis clearly reported: **2**	**11**

Study and number of participants	Selection bias	Control group	Associative factors (bias where retrospective reporting or not self-report)	Confounds	Outcomes	Statistics	Quality Total
5a. Davies (1988) (n=55)	Small sample – clinics and support organisation: **1**	No control group: **0**	Closeness index (retrospective report by parents): **0**	No pretesting, no time effect, but in original study only one sibling used: **1**	Outcomes on validated measure, but no third party: **1**	Statistics described, but no clear hypothesis reported: **1**	**4**
5b. Worden et al. (1999) (n=75)	Sample derived from Davies (1988) above and McCown and Pratt (1985) below: **1**	Parentally bereaved siblings: **1**	Gender: Age:**1**	Cause of death accounted for Same number of months since loss Siblings from same family excluded: **1**	Outcome on validated measure, but no third party: **1**	Statistics described. Hypothesis not clearly reported: **1**	**6**
6.Demi and Gilbert (1987) (n=18)	Small sample – selection from professional contacts and vital statistics: **0**	No control group: **0**	Parents emotional distress – self-report and grief Parental functioning: **1**	Data from a sibling and a parent was analysed twice, no controls for gender or age: **0**	Outcomes on validated measure, but no third-party report and 9/19 response from 2 families: **0**	Statistics described. Hypothesis clearly reported: **2**	**3**
7. The Swedish Study **8a. Eilegard et al. (2013b) (n=174)**	All eligible siblings invited where Index Child had died of cancer in Sweden between 2000-2007 **2**	Control group matched for age, gender and place of residence: **2**	Losing a cancer 2-9 years previously: **2**	Siblings from same family included – but matching of control group assisted with confounds: **1**	Outcomes on validated measure, but no third-party report: **1**	Statistics described. Hypothesis clearly reported: **2**	**10**
7b. Wallin, Steineck, Nyberg, and Kreicbergs (2015) (n=174)	See above: **2**	No control group: **0**	Retrospective self-report of perception of communication with family, friends and healthcare professionals during illness after death**:** **0**	Siblings from same family included. No confounds adjusted for with: **0**	Outcomes on validated measure, but no third-party report: **1**	Statistics described. Hypothesis not clearly reported: **1**	**4**
7c. Eilertsen, Eilegard, Steineck, Nyberg, and	See above: **2**	No control group**: 0**	Retrospective self-report of perception of social support prior to and	Siblings from same family included. No confounds adjusted for: **0**	Outcomes on validated measure, but no third-party report: **1**	Statistics described. Hypothesis not	**4**

Study and number of participants	Selection bias	Control group	Associative factors (bias where retrospective reporting or not self-report)	Confounds	Outcomes	Statistics	Quality Total
Kreicbergs (2013) (n=174)			following the loss of a sibling: **0**			clearly reported: **1**	
7d. Sveen et al. (2014a) (n=174)	See above: **2**	No control group: **0**	Time since loss, age at the time of the study, age at the time of study, age at the death of sibling, gender, living with parents, employed, studying, educational level, dependent children, loss of significant person pre-or post-lost:**2**	Siblings from same family included. Regression accounted for many confounds: **1**	Outcomes on validated measure, but no third-party report: **1**	Statistics described. Hypothesis not clearly reported: **1**	**7**
7e. Lövgren, Jalmsell, Eilegård Wallin, Steineck, and Kreicbergs (2016) (n=174)	See above: **2**	No control group: **0**	Retrospective reports of siblings' experiences of death: **0**	Siblings from same family included. No confounds adjusted for: **0**	Outcomes on validated measure, but no third-party report: **1**	Statistics described. Hypothesis not clearly reported: **1**	**4**
8. Fletcher, Mailick, Song, and Wolfe (2013) (n=1621)	Large cohort study: **2**	Yes – non-bereaved siblings: **2**	Parent education, gender, age. Marital status and income: **2**	Controlled for famiy socioeconomic status Family structural factors (birth order and size of sibship). Siblings in same family included – all types of deaths included: **1**	Self-report, but measures of socio-economic factors: **1**	Statistics described. Hypotheses clearly reported: **2**	**10**
9 Gerhardt et al. (2012) (n=105)	Medium sample from cancer registries: **1**	Yes – matched for gender, race and closest birth date: **2**	Gender, age and closer in time to death: **2**	Only one eligible sibling in each family included – also matching in control group- no matching for	Social behaviour, peer acceptance and friendship- with third party	Statistics described. Hypotheses clearly reported: **2**	**10**

Study and number of participants	Selection bias	Control group	Associative factors (bias where retrospective reporting or not self-report)	Confounds	Outcomes	Statistics	Quality Total
				socio-economic sstatus or pre-testing: **1**	measures although not all validated: **2**		
10. Hogan (1988) (n=40)	Small sample, selection from self-help groups **- 0**	None – **0**	Time since death – **2**	No siblings from same family, other confounding factors taken into account – **1**	Grief reactions – by self-report – **1**	Statistics not well described. Hypothesis not clearly reported. – **0**	**4**
11. Hogan and Greenfield (1991) (n=127)	Large sample, selection from self-help groups – **1**	None – **0**	Time since death – Vulnerability vs resilience **1**	No pre-testing; unclear if more than one sibling from one famiy, females possibly over represented, but age range accounted for– **0**	Self-concept and bereavement scales - **1**	Statistics described. Hypothesis not clearly reported – **1**	**4**
12. Martinson et al. (1987) (n=29)	Small and sample from homecare for children dying of cancer**: 0**	None: **0**	Bereavement due to loss of sibling between 7-9 years previously: **1**	Large age range(8-18,unclear if included siblings from same family, but no account of gender or age at time of death: **1**	Self-report self-concept scale: **1**	Statistics described. Hypothesis not clearly reported: **1**	**4**
13. McCown and Pratt (1985) (n=66)	Small sample referred from paediatric inpatient, death support groups, funeral directors: **1**	None: **0**	Age, gender, length of illness, family size, funeral attendance, maternal status, time since death, before death behaviour, parent child-communication: **1**	Siblings from same family included in sample, no pre-testing but parents asked to report on previous behaviours: **1**	Behaviour reported sometimes by fathers and mothers: **1**	Statistics described. Hypothesis not clearly reported**: 1**	**5**
14. Morris et al. (2016) (n=62)	Sample recruited from paediatric care**: 1**.	None – **0**	Caregiver PTSD and depression symptoms and family functioning symptoms; parent gender: **1**	Siblings from same family not included, controls for age and gender, cause of	Outcomes measured by self-report: **1**	Statistics described. Hypothesis clearly reported: **2**	**7**

Study and number of participants	Selection bias	Control group	Associative factors (bias where retrospective reporting or not self-report)	Confounds	Outcomes	Statistics	Quality Total
				death and time since death No pre-testing: **2**			
15. Mulhern et al. (1983) (n=46)	Sample comprising two groups of homecare or hospital care (comparison groups): **1**	None – **0**	Sibling died within homecare programme or in hospital. Association between family environment and parental psychopathology with siblings – **1**	Siblings from same family included, no analyses on effect of age of sibling or time – **0**	Outcomes measured by self-report **– 1**	Statistics described. Hypothesis clearly reported – **2**	**5**
16. Pettle Michael and Lansdown (1986) (n=28)	Families identified through hospital records: **1**	None: **0**	Family adjustment – parental self-report: **1**	Siblings from same family included, large age range and length of time since Index Child's death: **0**	Outcomes reported by parental and teacher report, but no self-report: **1**	Statistics not described. Hypotheses not clearly reported: **0**	**3**
17. Rosenberg et al. (2015)(n=56)	Families identified where parents had previously taken part in research in relation to child dying of cancer**: 1**	None: **0**	Length of time since Index Child's death. Social support, perceptions of the illness experience in retrospect: **1**	Siblings from same family included. Siblings were asked to report on mood in retrospect, but analyses of co-variates diagnosis: **1**	Outcomes measured by self-report: **1**	Statistics described. Hypothesis not clearly reported: **1**	**5**
18. Stikkelbroek et al. (2016) *n*=	Cohort study (2230), but only 15 bereaved siblings: **1**	Matched – by non – bereaved group: **2**	Pre-loss mental health problems taken at time before loss: **1**	Considered effects of gender and social economic status: **1**	Outcomes measured by self-report: **1**	Statistics described. Hypothesis reported**:2**	**8**

Appendix 2

Appendix 2.1. Service Improvement Project. Journal Information

Extract from Author Guidelines for Submissions to the Mental Health Review Journal

Review process

Each paper is reviewed by the editor and, if it is judged suitable for this publication, it is then sent to at least two independent referees for double blind peer review.

Final submission

The author must ensure that the manuscript is complete, grammatically correct and without spelling or typographical errors. Before submitting, authors should check their submission completeness using the available Article Submission Checklist. Proofs will be emailed prior to publication.

Format

Article files should be provided in Microsoft Word format. LaTex files can be used if an accompanying PDF document is provided. PDF as a sole file type is not accepted, a PDF must be accompanied by the source file. Acceptable figure file types are listed further below.

Article Length

Articles should be between 4000 and 7000 words in length, except for literature reviews or review articles which have no word limit. This includes all text including references and appendices. Please allow 350 words for each figure or table.

Article Title

A title of not more than eight words should be provided.

Author details

All contributing authors' names should be added to the ScholarOne submission, and their names arranged in the correct order for publication. •Correct email addresses should be supplied for each author in their separate author accounts

•The full name of each author must be present in their author account in the exact format they should appear for publication, including or excluding any middle names or initials as required

•The affiliation of each contributing author should be correct in their individual author account. The affiliation listed should be where they were based at the time that the research for the paper was conducted

Biographies and acknowledgements

Authors who wish to include these items should save them together in an MS Word file to be uploaded with the submission. If they are to be included, a brief professional biography of not more than 100 words should be supplied for each named author.

Research funding

Authors must declare all sources of external research funding in their article and a statement to this effect should appear in the Acknowledgements section. Authors should describe the role of the funder or financial sponsor in the entire research process, from study design to submission.

Structured Abstract

Authors must supply a structured abstract in their submission, set out under 4-7 sub-headings (see our "How to... write an abstract" guide for practical help and guidance):
•Purpose (mandatory)

•Design/methodology/approach (mandatory)

•Findings (mandatory)

•Research limitations/implications (if applicable)

•Practical implications (if applicable)

•Social implications (if applicable)

•Originality/value (mandatory)

Maximum is 250 words in total (including keywords and article classification, see below).

Authors should avoid the use of personal pronouns within the structured abstract and body of the paper (e.g. "this paper investigates..." is correct, "I investigate..." is incorrect).

Keywords

Authors should provide appropriate and short keywords in the ScholarOne submission that encapsulate the principal topics of the paper (see the How to... ensure your article is highly downloaded guide for practical help and guidance on choosing search-engine friendly keywords). The maximum number of keywords is 12. Whilst Emerald will endeavour to use submitted keywords in the published version, all keywords are subject to approval by Emerald's in house editorial team and may be replaced by a matching term to ensure consistency.

Article Classification

Authors must categorize their paper as part of the ScholarOne submission process. The category which most closely describes their paper should be selected from the list below. Research paper. This category covers papers which report on any type of research undertaken by the author(s). The research may involve the construction or testing of a model or framework, action research, testing of data, market research or surveys, empirical, scientific or clinical research.

Headings must be concise, with a clear indication of the distinction between the hierarchies of headings. The preferred format is for first level headings to be presented in bold format and subsequent sub-headings to be presented in medium italics.

Notes/Endnotes

Notes or Endnotes should be used only if absolutely necessary and must be identified in the text by consecutive numbers, enclosed in square brackets and listed at the end of the article.

Figures

All Figures (charts, diagrams, line drawings, web pages/screenshots, and photographic images) should be submitted in electronic form. All Figures should be of high quality, legible and numbered consecutively with Arabic numerals. Graphics may be supplied in colour to facilitate their appearance on the online database. •Figures created in MS Word, MS PowerPoint, MS Excel, Illustrator should be supplied in their native formats. Electronic figures created in other applications should be copied from the origination software and pasted into a blank MS Word document or saved and imported into an MS Word document or alternatively create a .pdf file from the origination software.

Tables

Tables should be typed and included in a separate file to the main body of the article. The position of each table should be clearly labelled in the body text of article with corresponding labels being clearly shown in the separate file. Ensure that any superscripts or asterisks are shown next to the relevant items and have corresponding explanations displayed as footnotes to the table, figure or plate.

References

References to other publications must be in Harvard style and carefully checked for completeness, accuracy and consistency. This is very important in an electronic environment because it enables your readers to exploit the Reference Linking facility on the database and link back to the works you have cited through CrossRef.

Appendix 2.2. SIP. University Ethics Approval Confirmation

psychology-ethics
Fri 11/03/2016 13:32

Mark as unread

To: ☐ Lucy Fiddick;

● You replied on 20/04/2016 09:48.

MessageHeaderAnalyzer + Get more apps

Dear Lucy Fiddick

Reference Number 15-248

Thank you for satisfactorily attending to those amendments. I can now confirm that you have full ethical approval for your study.

Best wishes with your research,
Dr Michael J Proulx
Chair, Psychology Research Ethics Committee

Appendix 2.3. SIP. AWP R& D approval

Avon and Wiltshire NHS
Mental Health Partnership NHS Trust

Avon and Wiltshire Mental Health Partnership AWP Trust
AWP Quality Academy
Fromeside- East Wing
Manor Road
Fishponds
BS16 2EW

0117 378 4238/ 07825 725296

Lucy Fiddick,

Clinical Psychologist in Training,

lf431@bath.ac.uk

Date: 23rd February 2016

Dear Lucy

A study of decision making processes involved in referral to highlight and address problems in the referral process.

AWP Reference: E2016.007 Fiddick

This letter is to confirm that your evaluation is now approved and also provides you with our reference number.

If you do need any further support or information, please contact us using the contact details above, quoting our reference number for your study.

The importance of disseminating all evaluation work cannot be over emphasised. It is only by sharing our learning that we can improve services across AWP. For this reason, the findings of all evaluation work should be reported to the Evaluation team via email. The team will champion the results of service evaluations, and work with evaluators to ensure those results are disseminated and acted upon, and that the results of evaluations are reflected in future service delivery. The team will also work with evaluators to produce publications for the public domain.

I very much look forward to receiving the results of your evaluation in due course.

Yours sincerely,

Janet Brandling

Appendix 2.4. SIP. Participant Information Sheet and Consent Form

NHS
Avon and Wiltshire Mental Health Partnership
NHS Trust

Participant Information Sheet (recovery navigators)

We would like to invite you to take part in our evaluation. Before you decide it is important for you to understand why the evaluation is being done and what it will involve. Please take time to read the information and discuss it with others if you wish. If anything is unclear or you have any questions, please ask us.

Title of the study

A study of decision making processes involved in referral to psychological services within secondary mental health.

What is the purpose of the study

We know that within Bristol, service users gain access to psychological therapies when the referrer makes a request for input from complex psychological interventions (CPI) on an online form. This means that care co-ordinators and recovery team staff are important in enabling service users to access psychological therapies. We are interested in discussing with people who refer to CPI how they come to a decision to refer people to CPI and why they might decide not to refer a SU to CPI. This evaluation therefore seeks to answer the question: What do referrers within RS identify as the factors that influence their decisions to refer to psychological services?

Do I have to take part?

It is completely your choice as to whether you wish to take part or not. There are no consequences for not taking part on your employment with AWP, and similarly there will be no consequence to employment if you decide to withdraw.

What will I be asked to do if I take part?

Recovery Navigators who work within the central Recovery Team are invited to take part in a focus group to discuss their decision-making process in referring to CPI. The focus group will run for an hour and will be audio or video recorded and transcribed to identify any themes. On completion of the project a summary of the findings will be made available to all members of staff working in the Recovery and Assessment Team.

The focus group discussion will take place at 2:15 pm on Thursday 21 April 2016 and you will be offered a late free sandwich lunch.

Will what I say be kept confidential?

Everything said in the interview will be anonymised and so no one will be able to find out what you have said in the interview. People participating in the focus group will be asked to abide by ground rules and not discuss what has individuals have spoken about outside the focus group.

All information will be kept confidential and will conform to the Data Protection Act of 1998 with respect to collection, storage and destruction of data. All paper-based and electronic information will be locked away and will be protected with a password. All data will have your name removed so you will not be able to be formally identified. This means that the only person to have access to identifiable information will be Lucy Fiddick. Access to other information will be restricted to James Gregory and Lucy Fiddick.

To make sure all valuable information is collected during the focus group the evaluation project will be using either a digital audio recorder video recorder. Recordings will be destroyed on the completion of the project.

The results will be fed back to AWP and possibly reported in mental health journals. The findings will also contribute to Lucy Fiddick's doctorate in clinical psychology. No one will be identified in any of these reports and publications.

What if there is a problem?

If you have any concerns or wish to complain about any aspect of the evaluation project then you should initially contact the researchers, Lucy Fiddick, Dr James Gregory or Dr Falguni Nathwani. Alternatively you can contact your line manager or team manger within

the Recovery and Assessment Team. If you decide you no longer wish to participate in the project, you can opt out.

What to do next if I am interested?

If you wish to take part please email Lucy Fiddick. If there are too many volunteers, participants will be chosen at random. You will also be asked to complete the attached consent form on the day of the focus group interview. You can contact James, Lucy or Falguni at any time to ask questions

Lucy Fiddick: lucy.fiddick@nhs.net (07811 321175)

Lucy's supervisor

James Gregory: j.d.gregory@bath.ac.uk (01225) 386120

Field supervisor of the project

Falguni Nathwani: falguni.nathwani@nhs.net (0117) 954 1954

NHS
Avon and Wiltshire Mental Health Partnership
NHS Trust

My name is Lucy Fiddick and I am a clinical psychologist in training based at the University of Bath.

I am completing a Doctorate in Clinical Psychology, and my work is being supervised by Dr James Gregory. James and I will be the only two people who will have access to any identifiable information from the questionnaires and the focus group.

The information sheet explains the following to you:

- The nature and the purpose of this study.
- Why you have been asked to participate in the study.
- What will be required of you as part of the study.
- That the discussion in the focus group will be audio recorded in order for it to be transcribed.
- That you have the right to withdraw from the study at any point you wish to and that you can request for information that has already been provided to be destroyed.
- That it will not affect your employment with AWP in any way if you do not participate in the study or withdraw from the study.

	Please tick
I have had the above explained to me and I agree to participate in the study.	
I agree for Lucy Fiddick and her supervisor to have access to the information produced from my responses for the purposes of this study.	
I agree for the focus group to be recorded.	

Consent

I **agree** to participate in the study
Signed…… Date……

I **do not agree** to participate in the study

Signed …….. Date …….

When completed, one copy for the participant, one copy for the researcher's files.

Appendix 2.5. SIP. Power point presentation of findings

WHY DO RECOVERY TEAM MEMBERS REFER FOR PSYCHOLOGICAL INPUT

How recovery team employees make decisions to refer: a qualitative study – Lucy Fiddick, Trainee Clinical Psychologist,

HOW DO REFERRERS DECIDE WHO TO REFER TO PSYCHOLOGICAL THERAPIES?

BACKGROUND

Psychological therapy is an effective treatment for a range of mental health problems including depression, psychosis and personality disorders (NICE, 2005, 2009a, b, 2014).

It is also the preferred treatment for people with mental health problems for conditions including depression and anxiety (Prins et al., 2008; van Schaik et al., 2004).

Psychological services like CPI can make decisions about **what** therapies people should access, but referrers are the initial gate keepers for **who** accesses psychological input.

EVIDENCE BASE ABOUT DECISIONS TO REFER TO PSYCHOLOGY

Martin (1999) identified factors influencing nurses' decision-making in mental health nursing. He identified three inter-related concepts:

Service users' personality

Self –the nurse's own attitudes, beliefs, values and changing emotional states.

Social system -the structure of the healthcare system and the physical and social environment impacted on decision-making.

MATRIX MODEL OF CLINICAL JUDGEMENT MARTIN (1999)

WHO ARE THE GATEKEEPERS OF BRISTOL CPI?

Assertive engagement service, community assessment and recovery service, early intervention in psychosis service, crisis service and inpatient service can refer to CPI.

EVIDENCE BASE ABOUT DECISIONS TO REFER TO PSYCHOLOGY

Martin's (1999) model maps on to findings of studies considering how decisions to refer to psychology have looked at GPs.

Qualities of the service user - those who are more "high risk" are referred (Sigel & Lieber, 2004) those who will benefit most and patients who ask for a referral (Stavrou, Cape and Sorkes, 2009).

GP's beliefs – i.e. the **self** - GPs who are psychologically minded refer more (Fitch, Dow, Solmes & Gray, 2008). Conversely they refer more when a service user evokes negative emotional responses in GPs (Knight, 2003)

Social structure – limited resources within therapy mean that GPs prioritise those who would use the referral well (Stavrou, Cape and Sorkes, 2009)

AIM AND RATIONALE

AIM: To understand how recovery workers decide how they refer into psychological therapies and consider what could be done to help them in their decision making

RATIONALE: Recovery navigators and Recovery Practitioners are the gatekeepers to psychology in CPI – they decide who is referred for psychological therapy and who obtains indirect psychological input through stream meetings, formulation sessions, team meetings and one to ones AND who does not.

HOW DO RECOVERY WORKERS MAKE DECISIONS TO REFER TO CPI?

Focused on how recovery workers make decisions to refer because the majority of referrals derive from Recovery Service.

Recovery Practitioners (RPs) and Recovery Navigators (RNs) in the central Bristol team were invited to take part in the study by email and announcement at the team meeting.

METHOD – INTERVIEWEES

HOW DO RECOVERY WORKERS MAKE DECISIONS TO REFER TO CPI?

Interviews conducted individually or in small groups with RPs and RNs interviewed separately to encourage participants to speak freely.

6 RPs and 5 RNs were interviewed in six separate interviews in May and June 2016.

All from Central Bristol.

METHOD - INTERVIEWS

Interviews were conducted using a semi-structured interview using prompts relating to Martin matrix to understand how the following contributed to decisions made:

Social structure

Service user qualities

Participant's own attitudes and beliefs

METHODS – QUALITATIVE ANALYSIS

Interviews were transcribed and analysed using thematic analysis (Braun and Clarke (2006)

Conducted from a realist perspective reflecting an assumption that words conveyed meaning.

A deductive approach was used to ensure that the research question was addressed and so codes relating to the social structure, the SU and participants' beliefs were identified and guided the process of generating themes.

THEMES

3 main themes–
Organizational structure,
SU qualities and
Beliefs about psychology

4th minor theme –
Improvements

RESULTS- FINDINGS FROM ANALYSIS

ORGANISATIONAL STRUCTURE

External pressures
Uncertainty about criteria
Limits in resource
Indirect psychological input

ORGANISATION STRUCTURE AND WIDER SOCIAL SYSTEMS – EXTERNAL PRESSURES

RN4: But sometimes there is pressure as well from sort of doctors' reviews. So there have been some [psychiatrists] who say, "Yes, psychology, psychology....." No one here at the moment, but there has been almost promises to the client.
But it's not like we refer to psychology and they say yes. It takes time and there's criteria.

ORGANISATION STRUCTURE AND WIDER SOCIAL SYSTEMS- LIMITS IN RESOURCES

RP5: For me yeah, because I just think, I want to make sure I'm getting the most prioritised people into it and want to make sure if I am getting somebody into it, it has to be somebody who really needs it. If I get that wrong I kind of think like "Oh, that's an opportunity missed.

ORGANISATION STRUCTURE AND WIDER SOCIAL SYSTEMS -UNCERTAINTY ABOUT CRITERIA TO BE ACCEPTED

RN3: I think myself and a lot of recovery navigators are still a bit hazy about the criteria for acceptance and whether someone is going to get on to CPI or not.

RP6: I guess nobody has ever said... I've made up in my mind, it's entirely me whose made up who's going to benefit from psychology.

ORGANISATION STRUCTURE AND WIDER SOCIAL SYSTEMS- LIMITS IN RESOURCES

RP2: … And I think before, I was just thinking, unless someone is engaging really well – it's a real clear need…Yeah, I probably perhaps just wouldn't refer because it perhaps wouldn't get taken up. Whereas I might now be more willing to think, "Oh perhaps I will refer this person because perhaps they have more capacity."

ORGANISATION STRUCTURE AND WIDER SOCIAL SYSTEMS -INDIRECT PSYCHOLOGICAL INPUT

RP3: I am not wasting the time doing a referral if they are not going to take them up. I don't have time to do referrals for the sake of it – none of us do. Honestly. So that's probably not the right way, but I will stick to actually talking to a human being.

SU QUALITIES

Four themes:
SU's view of therapy
Stable enough to engage
Psychological insight
Ready of not to change

ORGANISATION STRUCTURE AND WIDER SOCIAL SYSTEMS -INDIRECT PSYCHOLOGICAL INPUT

RP2: I would bring it up in stream. Probably, because if you are referring somebody for psychology then you have got concerns about their wellbeing anyway and what's best for them so I would be talking to them at stream meeting anyway.

SU QUALITIES- SU'S VIEW OF THERPAY

RN2: I have not come across someone where they haven't asked and I've thought, "They really need it yet." So that's where I am, that's pretty much how I do it. They probably say, "I could probably do with some therapies or talking therapies or whatever else they ask for, I will then talk to CPI.

SU -STABLE ENOUGH TO ENGAGE

RP2: Well, there are people that we find it hard to engage with and that's us going out to houses, GP surgeries, community centres. So, that's us being very proactive I guess… So I guess, it's that those types of service users, I really wouldn't refer to psychology because I know how psychology works

SU -READY OR NOT TO CHANGE

RN2: *I think it's the individuals who want the most change who get the most out of it. That would be my sort of feeling.*

SU- PSYCHOLOGICAL INSIGHT

RP2: *They need insight to begin with… Yeah. An awareness that shifting something in them can make a change, rather than wanting the world to change, or other people to change.*

BELIEFS ABOUT AND ATTITUDES TO PSYCHOLOGY

Most people can benefit

CPI is not always the answer

Psychological input for high risk Sus

CPI has the answers

PARTICIPANTS' BELIEFS AND ATTITUDES TO PSYCHOLOGY – MOST PEOPLE CAN BENEFIT

RN4: *In an ideal world quite a lot of people [would benefit from psychology] really. That's why they come into the service because they need support with their mental health and their emotions.*

PARTICIPANTS BELIEFS AND ATTITUDES TO PSYCHOLOGY – WHEN SUS ARE HIGH RISK

RN4: So they might not fit the criteria for psychology, but it would be really helpful to have psychology discussion because you are really struggling.

IMPROVEMENTS

CPI to provide more training

The bloody form

IMPROVEMENTS – THE BLOODY FORM

RN2: *The form just plays me up all the time, but apart from that, it's all right…. I can't submit it because it won't allow me to put it in properly for some reason. There is just one piece of it which I can't put it in. It won't let me put it in the box*

IMPROVEMENTS – CPI TO PROVIDE MORE TRAINING

RP3: Who are *the most suitable people, are there particular diagnoses? Are there particular difficulties or whatever you want to call them.*

CONCLUSIONS

CONCLUSIONS

- All practitioners said that they believed psychology could benefit people in the right circumstances.

-Many practitioners said that it was easier to access psychology than previously.

- People used stream meetings, formulation sessions and team meetings for indirect psychological input – although some people found stream meetings a difficult place to talk.

- It was upbeat! Most practitioners felt things were getting better.

TENTATIVE RECOMMENDATIONS

RNs and RPs to be trained about the referral process to CPI

Further limited training on psychological therapies and who might benefit – including information about what is available for people who fall into the "gap" between services?

Correct that glitch in the form (this may already have happened)

Circulate availability sheets of what therapists are currently doing within CPI and whether they have capacity

CONCLUSIONS – BUT!

-There was lots of variation in views. Some practitioners purely led by external pressures or desires of service users. Others had strong views about who would not be suitable for therapy and did not ask psychologists even if the service user was asking.

- Often people spoke about feeling that the process of who could bene being accepted into psychology was mysterious.

- RPs and RNs also talked about there being a gap within services between people who are too risky for IAPT, but "too well for referral to psychological services – these are the people who are not talked about.

WIDER CONSIDERATIONS

Consider the gap in provision between IAPT and secondary mental health – addressed through joint ventures (e.g. steps programmes between off the record and IAPT)

Psychologists as care co-ordinators?

Would be good to have a better understanding of referral decisions of other professionals eg psychiatrists, general practitioners and primary care liaison?

YOUR THOUGHTS AND COMMENTS!

Appendix 3

Appendix 3.1. MRP. Journal information

Journal of Reproductive and Infant Health Psychology

5 year impact factor: 1.286

About the journal

Journal of Reproductive and Infant Psychology is an international, peer reviewed journal, publishing high-quality, original research. Please see the journal's Aims & Scope for information about its focus and peer-review policy.

Please note that this journal only publishes manuscripts in English.

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All authors submitting to medicine, biomedicine, health sciences, allied and public health journals should conform to the Uniform Requirements for Manuscripts Submitted to Biomedical Journals, prepared by the International Committee of Medical Journal Editors (ICMJE).

Word limits

Please include a word count for your paper.
A typical article for this journal should be no more than 3500 words; this limit does not include tables, references, figure captions, endnotes.

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Please refer to these style guidelines when preparing your paper, rather than any published articles or a sample copy.

Please use British -ise spelling style consistently throughout your manuscript.

Please use single quotation marks, except where 'a quotation is "within" a quotation'. Please note that long quotations should be indented without quotation marks.

Formatting and templates

Papers may be submitted in any standard format, including Word and LaTeX. Figures should be saved separately from the text. To assist you in preparing your paper, we provide formatting templates.

A LaTeX template is available for this journal.

Word templates are available for this journal. Please save the template to your hard drive, ready for use.

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References

Please use this reference style guide when preparing your paper. An EndNote output style is also available to assist you.

Checklist: what to include

1. **Author details**. Please ensure everyone meeting the International Committee of Medical Journal Editors (ICMJE) requirements for authorship is included as an author of your paper.Please ensure everyone meeting the International Committee of Medical Journal Editors (ICJME) requirements for authorship is included as an author of your paper.Please include all authors' full names, affiliations, postal addresses, telephone numbers and email addresses on the title page. Where available, please also include ORCID identifiers and social media handles (Facebook, Twitter or LinkedIn). One author will need to be identified as the corresponding author, with their email address normally displayed in the article PDF (depending on the journal) and the online article. Authors' affiliations are the affiliations where the research was conducted. If any of the named co-authors moves affiliation during the peer-review process, the new affiliation can be given

as a footnote. Please note that no changes to affiliation can be made after your paper is accepted. Read more on authorship.

2. A structured **abstract** of no more than 250 words. A structured abstract should cover (in the following order): Objective, Background, Methods (to include design and participants), Results, and Conclusion Read tips on writing your abstract.
3. **Graphical abstract** (Optional). This is an image to give readers a clear idea of the content of your article. It should be a maximum width of 525 pixels. If your image is narrower than 525 pixels, please place it on a white background 525 pixels wide to ensure the dimensions are maintained. Save the graphical abstract as a .jpg, .png, or .gif. Please do not embed it in the manuscript file but save it as a separate file, labelled GraphicalAbstract1.
4. You can opt to include a **video abstract** with your article. Find out how these can help your work reach a wider audience, and what to think about when filming.
5. 5-6 **keywords**. Read making your article more discoverable, including information on choosing a title and search engine optimization.
6. **Funding details**. Please supply all details required by your funding and grant-awarding bodies as follows:
   For single agency grants: This work was supported by the[Funding Agency] under Grant [number xxxx].
   For multiple agency grants: This work was supported by the [funding Agency 1]; under Grant [number xxxx]; [Funding Agency 2] under Grant [number xxxx]; and [Funding Agency 3] under Grant [number xxxx].
7. **Disclosure statement**. This is to acknowledge any financial interest or benefit that has arisen from the direct applications of your research. Further guidance on what is a conflict of interest and how to disclose it.
8. **Geolocation information.** Submitting a geolocation information section, as a separate paragraph before your acknowledgements, means we can index your paper's study area accurately in JournalMap's geographic literature database and make your article more discoverable to others.
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10. **Figures.** Figures should be high quality (1200 dpi for line art, 600 dpi for grayscale and 300 dpi for color, at the correct size). Figures should be saved as TIFF, PostScript or EPS files. More information on how to prepare artwork.
11. **Tables.** Tables should present new information rather than duplicating what is in the text. Readers should be able to interpret the table without reference to the text. Please supply editable files.
12. **Equations**. If you are submitting your manuscript as a Word document, please ensure that equations are editable. More information about mathematical symbols and equations.
13. **Units.** Please use SI units (non-italicized).

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Disclosure statement

Please include a disclosure of interest statement, using the subheading "Disclosure of interest." If you have no interests to declare, please state this (suggested wording: *The authors report no conflicts of interest*). For all NIH/Wellcome-funded papers, the grant number(s) must be included in the disclosure of interest statement. Read more on declaring conflicts of interest.

Clinical Trials Registry

In order to be published in a Taylor & Francis journal, all clinical trials must have been registered in a public repository at the beginning of the research process (prior to patient enrolment). Trial registration numbers should be included in the abstract, with full details in the methods section. The registry should be publicly accessible (at no charge), open to all prospective registrants, and managed by a not-for-profit organization. For a list of registries that meet these requirements, please visit the WHO International Clinical Trials Registry Platform (ICTRP). The registration of all clinical trials facilitates the sharing of information among clinicians, researchers, and patients, enhances public confidence in research, and is in accordance with the ICMJE guidelines.

Complying with ethics of experimentation

Please ensure that all research reported in submitted papers has been conducted in an ethical and responsible manner, and is in full compliance with all relevant codes of experimentation and legislation. All papers which report *in vivo* experiments or clinical trials on humans or animals must include a written statement in the Methods section. This should explain that all work was conducted with the formal approval of the local human subject or animal care committees (institutional and national), and that clinical trials have been registered as legislation requires. Authors who do not have formal ethics review committees should include a statement that their study follows the principles of the Declaration of Helsinki.

Consent

All authors are required to follow the ICMJE requirements on privacy and informed consent from patients and study participants. Please confirm that any patient, service user, or participant (or that person's parent or legal guardian) in any research, experiment, or clinical trial described in your paper has given written consent to the inclusion of material pertaining to themselves, that they acknowledge that they cannot be identified via the paper; and that you have fully anonymized them. Where someone is deceased, please ensure you have written consent from the family or estate. Authors may use this Patient Consent Form, which should be completed, saved, and sent to the journal if requested.

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Please confirm that all mandatory laboratory health and safety procedures have been complied with in the course of conducting any experimental work reported in your paper. Please ensure your paper contains all appropriate warnings on any hazards that may be involved in carrying out the experiments or procedures you have described, or that may be involved in instructions, materials, or formulae.

Please include all relevant safety precautions; and cite any accepted standard or code of practice. Authors working in animal science may find it useful to consult the International Association of Veterinary Editors' Consensus Author Guidelines on Animal Ethics and Welfare and Guidelines for the Treatment of Animals in Behavioural Research and Teaching. When a product has not yet been approved by an appropriate regulatory body for the use described in your paper, please specify this, or that the product is still investigational.

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If you are submitting in LaTeX, please convert the files to PDF beforehand (you may also need to upload or send your LaTeX source files with the PDF).

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On acceptance, we recommend that you keep a copy of your Accepted Manuscript. Find out more about sharing your work.

Appendix 3.2. MRP. Ethics approval

psychology-ethics

Fri 28/10/2016 12:54

Inbox

Mark as unread

Dear Lucy,

Re: Application 16-229

I am happy to approve your amended ethics application via Chair's Action. Thank you very much for making these changes and sending through the amended documents for our files. The code 16-229 is your ethics code to be used as proof of ethical approval.

Best of luck with your data collection,
Dr. Nathalia Gjersoe
Chair, Psychology Ethics Committee

Appendix 3.3. MRP. Information Sheet

Information Sheet

Emotional reactions to stillbirth and neonatal death of a baby

We would like to invite you to take part in our research study. Before you decide whether or not to take part, it is important to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully and discuss it with others if you wish. If you have any questions or if anything is not clear please contact Lucy Fiddick on perinatal-research@bath.ac.uk.

Reasons for this study

Stillbirth and the death of the baby before they are 28 days old (sometimes called perinatal death) are among the most heart-breaking experiences that women and their partners can go through. Each person has a unique experience of their bereavement and so will have different grieving experiences. As time passes, some people will find their grief more manageable while, for others, grief lasts longer and is more distressing.

We are interested in how different thoughts and feelings may link to grief. We are particularly interested in whether women have different feelings if at the time their baby died, they already had a living child or children or if they had not had a living child. We hope the outcomes of the research project can improve our understanding of grief following perinatal deaths and improve the help and support offered.

Who can take part in the study?

The following groups of women over the age of 18 are invited to take part in our study:

- Women who experienced stillbirth (at 24 weeks of pregnancy or later) or the death of their baby before he or she is 28 days old, between 13 and 36 months ago. We will divide this group into two sub-groups:
  - women who had at least one living child at the time of the death of their baby.
  - women who did not have a living child at the time of the death of their baby.
- Women who have had a surviving baby between 13 and 36 months ago and have never experienced the death of a baby before he or she was 28 days old. We need this group because we are interested in understanding similarities and differences between women who have experienced perinatal death and those who have not.

Women who cannot take part

We recognise that women have very different experiences of stillbirth or their baby dying which are important to understand, but we are only able to consider certain groups of women and types of perinatal death in this research. For this reason, we are not inviting the following groups to take part:

- women who have had a late termination of a pregnancy because of medical reasons.
- women who have had a multiple pregnancy in which one baby was either stillborn or did not survive beyond 28 days.
- women whose baby died more recently than 13 months ago or more than 36 months ago.
- women whose baby died before 24 weeks of pregnancy
- women who are under the age of 18.
- women who cannot read English will not be able to take part in the study (because we are not able to translate the questionnaires).

What do I have to do if I take part?

- If you decide to take part, you can choose to either:
- complete the questionnaires online; or
- complete the survey by telephone with the researcher.

Whichever method you choose, you will answer the same questions about your mental well-being and also about your thoughts and feelings now and in different scenarios. If your baby died, we will also ask you about your feelings of grief.

At the end of the study we will ask some brief questions about how you have found taking part in the research and if you have any comments. You can write or talk as little or as much as you would like and there are no wrong or right answers.

Completing the questionnaires either by telephone or online is expected to take between 30 and 45 minutes.

Online

If you choose to complete the survey online, we suggest you do the survey:

- at a time you can be quiet and will not be disturbed; and
- on a device you can easily type upon.

You will have the option of having a break and coming back to the research if you like by clicking on the "**finish later"** option at the bottom of each page. If you do this you can bookmark the survey or have the details emailed to you. Your responses will remain anonymous.

Telephone

If you think you would prefer to complete the survey on the telephone, please contact Lucy Fiddick on perinatal-research@bath.ac.uk. She will contact you within three working days and arrange a mutually convenient time to speak in the next few weeks.

Do I have to take part?

It is up to you to decide whether you wish to take part in the study or not. We will describe the study and then you can give consent if you are happy to. You can contact the research team on perinatal-research@bath.ac.uk if you would like to speak further before deciding whether or not you would like to take part.

Can I withdraw from the study?

You are free to stop responding to the questions at any time before you have completed the study your data will not be submitted and can be destroyed. Once you have completed the survey, the situation is different depending on whether you do the survey online or by telephone.

ONLINE: Your data will be anonymous and it will not be possible to identify and withdraw your data once you have finished the survey.

TELEPHONE: The researcher will keep a record of your identification number and you will be able to ask for your data to be destroyed up until the point the survey is closed and the data has been analysed.

Possible advantages/benefits from taking part?

We cannot promise the study will help you directly, but we hope your participation will improve our understanding of grief following perinatal death and improve the help and support offered. Previous research has shown that some people have found it helpful to take part in studies about their experiences of grief, and felt as though they were helping others by sharing their experiences.

On completion of the study, a summary of the findings of the study will be made available to participants who wish to receive them. You can request these by emailing us on perinatal-research@bath.ac.uk

Are there any disadvantages/risks from taking part?

Completing questionnaires will require you to think about thoughts and feelings that some people find more challenging. For those who have experienced perinatal death there will be some questions that ask about this. It is possible that you may find answering such questions upsetting. This is a normal reaction when thinking about difficult experiences and such feelings will normally subside

within a short time. It is important for you to understand you are not required to answer questions or share anything that you do not want to.

Some of the questions ask about your emotional wellbeing, but we will only be able to identify if you appear to be having difficulties if you complete the survey by telephone.

COMPLETING THE RESEARCH ONLINE: Your responses will be anonymous and so it will not be possible for the researcher to identify who you are and whether your answers suggest you may be feeling anxious or depressed. If you feel distressed after you have completed the questionnaires and would like the researcher to contact you can email her on perinatal-research@bath.ac.uk she will then arrange a time to speak on the telephone and be able to signpost you to organisations who might be able to help.

COMPLETING THE RESEARCH ON THE TELEPHONE: If you complete the survey by telephone, your responses will not be anonymous. Your researcher will let you know if your emotional wellbeing scores indicate that you may have depression or anxiety. She will also ask if you would like her to send you a letter stating the scores and suggest you make an appointment to see your general practitioner.

In both cases, if you are worried about your well-being, please make an appointment with your general practitioner (GP). If it is out of hours, you could contact the Samaritans on Helpline: 116 123

You could also contact other services that offer support (a list is at the end of this information sheet).

What if there is a problem?

We provide detailed information in part 2 of this information sheet about who to go to if you have any concerns or wish to complain about any aspect of the way you have been approached or treated as part of this study.

PART 2: INFORMATION SHEET

Will my taking part in this study be kept confidential?

Yes: although the situation is different depending on whether you complete the survey online or with a researcher on the telephone.

ONLINE: You will not be asked to provide your name or contact details if you take part in the study and so your data will be anonymous. If you email us to ask for information about the study, only the researchers will know your email address and this will be kept on a password protected memory stick.

TELEPHONE: The researcher will keep a record of your participant number together with details of your telephone number and email address. This will be kept on a password protected memory stick which will not hold your responses to the study.

What will happen to the results of the study?

This study is intended to form part of a doctoral thesis and to be submitted for publication in a relevant academic journal read by researchers and professionals working in this field. No individual participants will be identifiable in any written report resulting from this study. We will happily share the findings of this research with interested participants once they are available.

Who is organising and funding this research?

The project is being organised and funded by the University of Bath, as part of the Doctorate in Clinical Psychology for Lucy Fiddick.

Who has reviewed this project?

This research has received approval from the University of Bath Psychology Research Committee under reference 16-229.

What if there is a problem?

Every care will be taken to ensure your safety during the course of the study. If you have a concern about any aspect of this study, please contact the researchers on perinatal-research@bath.ac.uk. If you remain unhappy and wish to complain formally, you can contact Professor Jonathan Knight, Pro-Vice Chancellor for Research, c/o Dr Maria Wells, Executive Officer (Research), Vice-Chancellor's Office University of Bath, Claverton Down, Bath, BA2 7AY, United Kingdom (m.wells@bath.ac.uk).

If you would like to discuss the study further, you can contact:

Chief investigator: Lucy Fiddick, Clinical Psychologist in Training, University of Bath.

Academic supervisor: Dr Megan Wilkinson-Tough, University of Bath

Email: perinatal-research@bath.ac.uk

Organisations that offer support which could be helpful for you:

If after completing this survey you feel distressed and are worried about your mental wellbeing after completing this survey we recommend that you contact your general practitioner (GP) to discuss this. If it is out of hours, you could contact the following:

Samaritans
Available 24 hours a day to provide confidential emotional support for people who are experiencing feelings of distress or despair.
Website: samaritans.org.uk
Helpline: 116 123

Some other organisations that may be helpful are:

Child bereavement UK
Child bereavement UK is the UK's leading organisation that supports families when a baby or child of any age dies or is dying or when a child is facing bereavement.

Website: www.childbereavementuk.org
Helpline: 0800 02 888 40

SANDS (Stillbirth and Neonatal Death Charity)
Sands is a national charity that offers support to anyone affected by the death of a baby.
Website: www.uk-sands.org
Helpline: 020 7436 5881

Cruse Bereavement Care
Cruse Bereavement Care promotes the wellbeing of bereaved people and supports anyone bereaved by death to understand their grief and cope with loss.
Website: www.cruse.org.uk
Helpline: 0808 808 1677

Care for the Family
Care for the family is a national charity which aims to promote strong family life and to help those who face family difficulties, including bereavement.
Website: careforfamily.org.uk

Thank you for reading this information sheet.

Appendix 3.4. MRP. Consent form

Consent form

I have read and understood the information sheet for the study. I have been offered the opportunity to contact the research team to ask questions. * *Required*

- ○ Yes
- ○ No

I understand that my participation is voluntary and that my data will not be included in the research until I have submitted my responses at the end of the questions. I can therefore withdraw at any time whilst completing the questionnaires without my medical or legal rights being affected. Once I have submitted the completed questionnaires I will be unable to ask for this data to be withdrawn as it will not be possible to identify my anonymous responses. * *Required*

- ○ Yes
- ○ No

I understand that my responses are entirely anonymous and will be treated confidentially and any publication resulting from this work will report only data that does not identify me * *Required*

- ○ Yes
- ○ No

I am aware that this study involves completing questionnaires about my emotional wellbeing. * *Required*

- ○ Yes
- ○ No

I consent to participating in this study. * *Required*

- ○ Yes

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Appendix 3.5. MRP. Demographic questionnaire.

Demographic questions

About yourself

Please answer the following questions about yourself. This is to let us know whether the questionnaire has reached a wide range of different people and to check that you are eligible for the study.

What is your gender * *Required*

- Female
- Male
- Other

What is your age? * *Required*

- younger than 18 years
- 18 to 24 years
- 25 to 30 years
- 31 to 35 years
- 36 to 40 years
- 41 to 45 years
- 46 to 50 years
- 51 to 55 years
- 56 to 60 years
- Older than 18, but prefer not to say

Demographic Questionnaire

What is your ethnic group? Choose one option that describes your ethnic group best.

- White British
- Other White background
- Asian British
- Other Asian
- Black British
- Other Black
- Mixed/multiple ethnic groups
- Other

If you selected Other, please specify if you wish. *Optional*

What is the highest degree or level of schooling you have completed?

- No schooling.
- Left school before 16 with no qualifications
- GCSE qualifications or equivalent
- A-level or equivalent
- Degree qualification or above
- Prefer not to say
- Other

If you selected Other, please specify: *Optional*

What is your marital status?

- Single, never married
- Married or cohabiting with man
- Married/civil partnership or cohabiting with woman
- Divorced or separated

Have you experienced a stillbirth (at 24 weeks of pregnancy or later) or the death of a baby before he or she was 28 days old between 13 months and 3 years ago? * *Required*

- Yes
- No

Have you experienced the stillbirth (at 24 weeks of pregnancy or later) or death of a baby before he or she was 28 days old more recently than 13 months ago?

- Yes
- No

Was the death of your baby a result of a termination of your pregnancy for medical reasons?

- Yes
- No
- Prefer not to say

Did you have a multiple pregnancy (meaning that you were having twins, triplets or more children)? * *Required*

- Yes
- No
- Prefer not to say

Did any of your babies survive beyond 28 days after they were born? * *Required*

- Yes
- No

At the time your baby or babies died, did you have any other living children you have given birth to? *
Required

- ○ Yes
- ○ No

How many children did you have when your baby died and what ages are they now?

Please answer, if you answered yes to the question above. Do you consider your child or children to have health problem that will impact significantly on their life?

- ☐ Yes
- ☐ No

If you answered yes above, please give brief details if you would like.

At the time your baby or babies died, did your partner have any living children from a different relationship?

- ○ Yes
- ○ No
- ○ Prefer not to say

How many children your does your partner have and what are their ages now?

Are you pregnant now?

- Yes
- No
- Prefer not to say.

Have you given birth to a living baby or babies since your baby died?

- Yes
- No
- Prefer not to say

Appendix 3.6. MRP. Perinatal Grief Questionnaire

Perinatal Grief Scale

This part of the survey uses a table of questions, view as separate questions instead?

PRESENT THOUGHTS AND FEELNGS ABOUT YOUR BABY DYING Each of the items is a statement of thoughts and feelings which some people have concerning a loss such as yours. There are no right or wrong responses to these statements. For each item tick which best indicates the extent to which you agree or disagree with it at the present time. If you are not certain, use the "neither" category. Please try to use this category only when you truly have no opinion.

Please don't select more than 1 answer(s) per row.

Please select at least 1 answer(s).

	strongly disagree	disagree	neither disagree or agree	agree	strongly agree
I feel depressed	☐	☐	☐	☐	☐
I find it hard to get on with certain people	☐	☐	☐	☐	☐
I feel empty inside	☐	☐	☐	☐	☐
I can't keep up with my normal activities	☐	☐	☐	☐	☐
I feel a need to talk about the baby	☐	☐	☐	☐	☐
I am grieving for the baby	☐	☐	☐	☐	☐
I am frightened	☐	☐	☐	☐	☐
I have considered suicide since the loss	☐	☐	☐	☐	☐
I take medication for my nerves	☐	☐	☐	☐	☐
I very much miss my baby	☐	☐	☐	☐	☐
I feel I have adjusted well to the loss	☐	☐	☐	☐	☐
It is painful to recall memories of the loss	☐	☐	☐	☐	☐

I act upset when I think about the baby	☐	☐	☐	☐	☐
I cry when I think about him/her	☐	☐	☐	☐	☐
I feel guilty when I think about the baby	☐	☐	☐	☐	☐
I feel physically ill when I think about the baby	☐	☐	☐	☐	☐
I feel unprotected in a dangerous world since he/she died	☐	☐	☐	☐	☐
I try to laugh, but nothing seems funny anymore	☐	☐	☐	☐	☐
Time passes so slowly since the baby died	☐	☐	☐	☐	☐
The best part of me died with the baby	☐	☐	☐	☐	☐
I have let people down since the baby died	☐	☐	☐	☐	☐
I feel worthless since he/she died	☐	☐	☐	☐	☐
I blame myself for the baby's death	☐	☐	☐	☐	☐
I get cross at my friends and relatives more than I should	☐	☐	☐	☐	☐
Sometimes I feel like I need a professional counsellor to help me get my life back together again	☐	☐	☐	☐	☐
I feel as though I'm just existing and not really living since he/she died	☐	☐	☐	☐	☐

I feel so lonely since he/she died	☐	☐	☐	☐	☐
I feel somewhat apart and remote, even among friends	☐	☐	☐	☐	☐
It's safer not to love	☐	☐	☐	☐	☐
I find it difficult to make decisions since the baby died	☐	☐	☐	☐	☐
I worry about what my future will be like	☐	☐	☐	☐	☐
Being a bereaved parent means being a "Second Class Citizen"	☐	☐	☐	☐	☐
It feels great to be alive	☐	☐	☐	☐	☐

Submit and continue >

Appendix 3.7. MRP. TOSCA

TOSCA, Version 3

Below are situations that people are likely to encounter in day-to-day life, followed by several common reactions to those situations:

As you read each scenario, try to imagine yourself in that situation. Then indicate how likely you would be to react in each of the ways described. We ask you to rate all responses because people may feel or react more than one way to the same situation, or they may react in different ways at different times.

For an example see the question below.

This part of the survey uses a table of questions, view as separate questions instead?

You wake up early one Saturday morning. It is cold and rainy outside.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely or unlikely	4 a bit likely	5 very likely
a) You would telephone a friend to catch up on news.	☐	☐	☐	☐	☐
b) You would take the extra time to read the paper.	☐	☐	☐	☐	☐
c) You would feel disappointed that it's raining.	☐	☐	☐	☐	☐
d) You would wonder why you woke up so early.	☐	☐	☐	☐	☐

In the above example, I would rate ALL of the answers by ticking a number. I ticked a "1" for answer (a) because I wouldn't want to wake up a friend very early on Saturday morning - so it's not at all likely that I would do that. I ticked a "5" for answer (b) because I almost always read the paper if I have time in the morning (very likely). I ticked a "3" for answer (c) because for me it's about half and half. Sometimes I would be disappointed about the rain and sometimes I wouldn't --It would be depend on what I have planned. And I circled a "4" for answer (d) because I probably would wonder why I had awakened so early.

Please do not skip any items -- rate all responses.

This part of the survey uses a table of questions, view as separate questions instead?

You make plans to meet a friend for lunch. At five o'clock, you realise you have stood your friend up.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely or unlikely	4 likely	5 very likely
a) You would think: "I'm inconsiderate."	☐	☐	☐	☐	☐
b) You would think you should make it up to your friend as soon as possible.	☐	☐	☐	☐	☐
c)You would think: "My boss distracted me just before lunch."	☐	☐	☐	☐	☐

This part of the survey uses a table of questions, view as separate questions instead?

2. You break something at work and then hide it.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely or unlikely	4 a bit likely	5 very likely
a) You would think: "This is making me anxious. I need to either fix it or get someone else to."	☐	☐	☐	☐	☐
b) You would think about quitting.	☐	☐	☐	☐	☐
c) You would think: "A lot of things aren't made very well these days."	☐	☐	☐	☐	☐

At work, you wait until the last minute to plan a project, and it turns out badly.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely nor unlikely	4 a bit likely	5 very likely
a) You would feel incompetent.	☐	☐	☐	☐	☐
b) You would think: "There are never enough hours in the day."	☐	☐	☐	☐	☐
c) You would feel: "I deserve to be reprimanded for mismanaging the project.	☐	☐	☐	☐	☐

This part of the survey uses a table of questions, view as separate questions instead?

4. You make a mistake at work and find out a co-worker is blamed for the error.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely nor unlikely	4 a bit likely	5 very likely
a) You would think the company did not like the co-worker.	☐	☐	☐	☐	☐
b) You would keep quiet and avoid the co-worker.	☐	☐	☐	☐	☐
c) You would feel unhappy and eager to correct the situation.	☐	☐	☐	☐	☐

While playing around, you throw a ball and it hits your friend in the face.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely nor unlikely	4 a bit likely	5 very likely
a) You would feel inadequate that you can't even throw a ball.	☐	☐	☐	☐	☐
b) You would think maybe your friend needs more practice at catching.	☐	☐	☐	☐	☐
c) You would apologise and make sure your friend feels better.	☐	☐	☐	☐	☐

This part of the survey uses a table of questions, view as separate questions instead?

You are driving down the road, and you hit a small animal.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely nor unlikely	4 a bit likely	5 very likely
a) You would think the animal shouldn't have been on the road.	☐	☐	☐	☐	☐
b) You would think: "I'm terrible."	☐	☐	☐	☐	☐
c) You'd feel bad you hadn't been more alert driving down the road.	☐	☐	☐	☐	☐

7. You walk out of an exam thinking you did extremely well, then you find out you did poorly.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely nor unlikely	4 a bit likely	5 very likely
a) You would think: "The instructor doesn't like me."	☐	☐	☐	☐	☐
b) You would think: "I should have studied harder."	☐	☐	☐	☐	☐
c) You would feel stupid.	☐	☐	☐	☐	☐

This part of the survey uses a table of questions, view as separate questions instead?

While out with a group of friends, you make fun of a friend who's not there.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely nor unlikely	4 a bit likely	5 very likely
a) You would think: "I feel small...like a rat.	☐	☐	☐	☐	☐
b) You would think that perhaps that friend should have been there to defend himself/herself.	☐	☐	☐	☐	☐
c) You would apologise and talk about that person's good points.	☐	☐	☐	☐	☐

You make a big mistake on an important project at work. People were depending on you and your boss criticises you.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither likely or unlikely	4 a bit likely	5 likely
a) You would think your boss should have been more clear about what was expected of you.	☐	☐	☐	☐	☐
b) You would feel as if you wanted to hide.	☐	☐	☐	☐	☐
c) You would think: "I should have recognised the problem and done a better job."	☐	☐	☐	☐	☐

This part of the survey uses a table of questions, view as separate questions instead?

You are taking care of your friend's dog while they are on vacation and the dog runs away.

Please don't select more than 1 answer(s) per row.

	1 not likely	2 a bit unlikely	3 neither unlikely or likely	4 a bit likely	5 very likely
a) You would think, "I am irresponsible and incompetent."	☐	☐	☐	☐	☐
b) You would think your friend must not take very good care of her dog or it wouldn't have run away.	☐	☐	☐	☐	☐
c) You would vow to more careful next time.	☐	☐	☐	☐	☐

Appendix 3.8. MRP. PFQ-2

Personal Feelings Questionaire -2

Instructions: For each of the following listed feelings, indicate how common each feeling is for you.

0 = You never experience that feeling.
1 = You experience that feeling occasionally.
2 = You experience that feeling sometimes.
3 = You experience that feeling often.
4 = You experience that feeling all the time or almost all the time.

This part of the survey uses a table of questions, view as separate questions instead?

Read each item and the mark the appropriate answer in the space next to the word. Use the following scale to record your answers.

Please don't select more than 1 answer(s) per row.

	0 ever experience	1 experience occasionally	2 experience sometimes	3 experience often	4 experience all the time or almost all the time
Embarrassment	☐	☐	☐	☐	☐
Mild guilt	☐	☐	☐	☐	☐
Feeling ridiculous	☐	☐	☐	☐	☐
Worry about hurting or injuring someone	☐	☐	☐	☐	☐
Sadness	☐	☐	☐	☐	☐
Self-consciousness	☐	☐	☐	☐	☐
Feeling humiliated	☐	☐	☐	☐	☐
Intense guilt	☐	☐	☐	☐	☐
Euphoria	☐	☐	☐	☐	☐
Feeling "stupid"	☐	☐	☐	☐	☐
Regret	☐	☐	☐	☐	☐
Feeling "childish"	☐	☐	☐	☐	☐
Mild happiness	☐	☐	☐	☐	☐
Feeling helpless, paralysed	☐	☐	☐	☐	☐
Depression	☐	☐	☐	☐	☐
Feelings of blushing	☐	☐	☐	☐	☐
Feeling you deserve criticism for what you did	☐	☐	☐	☐	☐
Feeling laughable	☐	☐	☐	☐	☐
Rage	☐	☐	☐	☐	☐
Enjoyment	☐	☐	☐	☐	☐
Feeling disgusting to others	☐	☐	☐	☐	☐
Remorse	☐	☐	☐	☐	☐

Appendix 3.9. MRP. State Shame and Guilt Scale

SSG Scale

This part of the survey uses a table of questions, view as separate questions instead?

The following are some statements which may or may not describe how you are feeling right now. Please rate each statement using the 5 point scale below. Remember to rate each statement on how you are feeling *right at this moment*.

Please don't select more than 1 answer(s) per row.

	1 Not feeling this way	2	3 Feeling this way somewhat	4	5 Feeling this way very strongly
I feel good about myself.	☐	☐	☐	☐	☐
I want to sink into the floor and disappear.	☐	☐	☐	☐	☐
I feel remorse and regret.	☐	☐	☐	☐	☐
I feel worthwhile, valuable.	☐	☐	☐	☐	☐
I feel small.	☐	☐	☐	☐	☐
I feel tension about something I have done.	☐	☐	☐	☐	☐
I feel capable, useful.	☐	☐	☐	☐	☐
I feel like a bad person.	☐	☐	☐	☐	☐
I cannot stop thinking about something bad I have done.]	☐	☐	☐	☐	☐
I feel proud.	☐	☐	☐	☐	☐
I feel humiliated, disgraced.	☐	☐	☐	☐	☐
I feel like apologising, confessing.	☐	☐	☐	☐	☐
I feel pleased about something I have done.	☐	☐	☐	☐	☐
I feel worthless, powerless.	☐	☐	☐	☐	☐
I feel bad about something I have done.	☐	☐	☐	☐	☐

Appendix 3.10. Main Research Project. Questions about Research

Questions about completing these questionnaires

Has your experience of completing these questionnaires **at the moment** been:

- ☐ a very positive experience
- ☐ a positive experience
- ☐ neither a positive or negative experience
- ☐ a negative experience
- ☐ a very negative experience

Any comments

Do you believe that completing these questionnaires will be a positive or negative experience for you in the long-term?

- ☐ very positive
- ☐ positive
- ☐ neither positive or negative
- ☐ negative
- ☐ very negative

Any comments

Do you think you may contact someone (either your GP or another professional or support group) to speak about your baby dying as a result of taking part in this research?

- [ ] Yes
- [ ] No
- [ ] Not sure

Any comments?

Final question

Thank you for completing the survey. If you have any comments, please set these out below.